NEWBORN GENETIC SCREENING FOR TYPE I DIABETES FACTORS AFFECTING MATERNAL RISK PERCEPTION ANXIETY AND STUDY PARTICIPATION

By

STACY CARMICHAEL

A DISSERTATION PRESENTED TO THE GRADUATE SCHOOL
OF THE UNIVERSITY OF FLORIDA IN PARTIAL FULFILLMENT
OF THE REQUIREMENTS FOR THE DEGREE OF
DOCTOR OF PHILOSOPHY

UNIVERSITY OF FLORIDA

2003

ACKNOWLEDGMENTS

The completion of this project would not have been possible without my chair, Dr Suzanne Bennett Johnson, who has been a wonderful mentor and role model, and who possesses a thoughtful understanding of and a genuine concern for the well being and success of her students I am also grateful for the continuing moral and financial support, encouragement, and enthusiasm of Dr Desmond Schatz throughout my graduate career Further thank yous are sent to my other committee members for the generous donation of their time and thoughtful review of this dissertation, including Dr Stephen Boggs Dr Fonda Eyler and Dr Gary Geffken Data collection would not have been completed without the substantial efforts of Jennifer Walsh Adam Lewin, and Amy Baughcum, for which I am grateful My gratitude is further extended to Dr Rwanda LaToya Aker for providing support and humor during the final days of the dissertation completion process. I would also like to acknowledge Dr Robert Blodgett, my undergraduate mentor, for encouraging my initial pursuits in the field of psychology and pushing me to follow my dreams My son, Jaden Carmichael, has provided further inspiration to continue the pursuit of my doctoral degree Finally, I wish to thank my parents Ron and Donna Koser, and my best friend Mr Michael Walsh, for generous donations of love, encouragement, as well as physical financial and emotional support.

TABLE OF CONTENTS

Page

ACKNOWLEDGMENTS iii

ABSTRACT vi

CHAPTER

1 INTRODUCTION 1

2 REVIEW OF THE LITERATURE 5

- Complexities of Communicating Genetic Risk Information 6
  - Factors Influencing Risk Communication 6
  - Methods of Communicating Genetic Risk Information 11
- The Impact of Genetic Testing and Risk Communication 15
  - Cognitive Outcomes and Related Variables 17
  - Affective Response to Genetic Risk Communication 21
  - Behavioral Outcomes in Genetic Risk Communication Studies 25
  - Theoretical Models Predicting Effectiveness of Risk Communication 31
- Newborn Genetic Screening 33
  - Benefits and Risks of Newborn Genetic Screening 34
  - The Prediction and Prevention of Type I Diabetes 38

3 RATIONALE FOR AND PURPOSES OF THE STUDY 48

4 METHODS 52

- Participants 52
- Procedures 53
- Measures 56

5 RESULTS 62

- Demographic and Family Characteristics 62
- Accuracy of Maternal Risk Perception 66
- Maternal Anxiety in Response to Risk Notification 75
- Maternal Intent to Continue with PANDA Study 81

6 DISCUSSION 87

- Review of Findings and Conclusions 87

Maternal Understanding of Newborn Risk 87
Maternal Anxiety in Response to Risk Notification 91
Maternal Intent to Continue with PANDA Study 94
Limitations of the Current Study 100

Strengths of the Current Study and Final Comments 101

APPENDICES

A DIABETES RISK FACTORS 104

B PROCEDURAL FLOW OF STUDY 105

C SAMPLE NOTIFICATION INTERVIEW 106

D SAMPLE RISK INFORMATION LETTER, MONETARY EMPHASIS 110

E SAMPLE RISK INFORMATION LETTER, ALTRUISTIC EMPHASIS 111

F PHONE INTERVIEW SCRIPT FOLLOW-UP INTERVIEW 1 112

G PHONE INTERVIEW SCRIPT FOLLOW UP INTERVIEW 2 116

H PANDA GENERAL INFORMATION LETTER 119

LIST OF REFERENCES 120

BIOGRAPHICAL SKETCH 140

Abstract of Dissertation Presented to the Graduate School
of the University of Florida in Partial Fulfillment of the
Requirements for the Degree of Doctor of Philosophy

NEWBORN GENETIC SCREENING FOR TYPE I DIABETES: FACTORS AFFECTING MATERNAL RISK PERCEPTION, ANXIETY AND STUDY PARTICIPATION

By

Stacy Carmichael

December 2003

Chair: Suzanne Bennett Johnson
Major Department: Clinical and Health Psychology

Newborn genetic screening programs are available to the general population to assess a child's risk of developing Type I diabetes. The purpose of the following study was to evaluate the effect of a tailored risk information letter for a newborn genetic screening program for Type I diabetes on maternal accuracy of perceived risk, anxiety, and intentions to continue participation in a longitudinal diabetes-screening program.

Mothers who decided to obtain information regarding their child's genetic risk for developing Type I diabetes were randomized to a risk information letter or no-letter condition. Participants were interviewed approximately 1 month post risk notification and again 3.5 months later. Nearly 80% of the mothers were accurate in their recall of risk, with a tendency to underestimate risk over time. Maternal accuracy was related to higher maternal education, a longer notification phone call, receipt of a tailored risk information letter, and higher personal information seeking about their child's risk for developing diabetes. The tailored risk information intervention letter was also related to

maternal report of positive intentions to continue in the longitudinal screening study, as was having a family history of diabetes and identifying with an ethnic minority status. Underestimating the child's risk was also related to maternal intentions to not participate in the longitudinal screening study. No effect of the tailored risk information letter was observed on maternal anxiety. Rather, consistent predictors of maternal anxiety included having a higher infant risk, lower levels of dispositional optimism, and higher reports of maternal information-seeking.

Making the decision to participate in genetic screening studies is complicated and may be influenced by a variety of factors. In this population, no intervention to increase accuracy of risk perception subsequent to newborn genetic screening had been tested. It will be important to determine further what type of risk notification strategies work best to ensure that mothers understand the information they receive when deciding to participate in a longitudinal study of their child's risk for diabetes development.

CHAPTER I
INTRODUCTION

Over the past decade, science has made incredible biomedical progress that will have a significant impact on our lives in many ways that are currently unimaginable Only a mere fifty years after the discovery of the base pairs of deoxyribonucleic acid (DNA) in 1953, The Human Genome Project (HGP) which has had the goal of mapping and sequencing the entire human genome, completed a comprehensive foundation map of the human genome nearly two years ahead of schedule (Collins, Green, Guttmacher and Guyer 2003) The HGP has now embarked on a quest to identify, locate, and analyze the functional components located within our DNA, for the purposes of disease prediction and ultimately, disease prevention (Collins et al, 2003)

Genetic alterations can account for an estimated 5000 diseases as well as influence the development of thousands of others Genetic tests[1] are readily available that can identify disease or disorder-causing genes in those who are already ill, as well as those who are at risk for developing the disease at some point in their life and to inform reproductive decisions Some of these diseases which result from a complex interplay of genetic and environmental factors may be prevented, or the likelihood of disease reduced, by an individual's use of risk-reducing behaviors such as surveillance diet, exercise, or other preventative behaviors. However, the development of effective treatments is often a long and arduous process. Many of the diseases that will be identified in the future by genetic testing will not yet have effective interventions. In these diseases, it will be many years before knowledge of disease risk prior to disease onset will provide opportunities for risk reduction or disease prevention.

---

[1] Genetic testing refers to the use of "specific assays to determine the genetic status of an individual already suspected to be at high risk for a particular inherited condition because of family history or clinical symptoms while genetic screening involves the use of "various genetic tests to evaluate populations or groups of individuals independent of a family history of a disorder"(Institute of Medicine (IOM), 1994 p 4) However the terms "genetic testing" and "genetic screening are often used interchangeably as they will be in the following review

The surge of information and technology emanating from the HGP continues to grow exponentially. Tests will become more commonplace in every field of healthcare, and likely will be widely commercially available (Van McCrary, Allen, Moseley, Crandall, Ostrer, Curry, Dewar, & Nye, 1993). As it estimated each person carries from 5-30 alterations in DNA that could predispose the individual to the development or passing along of a genetically linked disease or disorder, no one will be exempt from the future of genetic testing.

The prospect is both exciting and frightening. The speed of technology is developing much faster than our understanding of the psychological, social, ethical, and legal implications that our genetic knowledge will bring. The National Human Genome Research Institute and the founders of the Human Genome Project, including Eugene Watson, were cognizant early on regarding the implications that information from the human genome would have on individuals and society (Peters, 1993). The Ethical, Legal, and Social Implications (ELSI) Research Program has functioned for more than a decade to examine privacy and fairness in the use and interpretation of genetic information, clinical integration of genetic technology, issues surrounding genetics research, and public and professional education about genetic testing (NHGRI, 2003). More questions than answers abound, including how individuals make the decision to be tested, how their perceptions of risk affect their health-behaviors; how individuals' decisions impact their psychological state, their family members and relationships, their finances, insurance or employment status, just to list a few

To address the concerns surrounding the implications of genetic testing several groups have published guidelines for conducting informed genetic testing as well as policy recommendations (e g , Alliance of Genetic Support Groups 1993, American Society of Human Genetics, 1996, Fuller et al. 1999 Moseley, Allen Van McCrary, Dewar, Crandall, & Ostrer, 1995 Project Policy Executive Summary, 2003) The Committee on Assessing Genetic Risks for the Institute of Medicine of the National Academy of Science for example published policy recommendations to guide responsible use of genetic testing programs for the purpose of maximizing effectiveness and minimizing problematic aspects of screening programs (IOM, 1994) The fundamental "four principle ' biomedical ethical approach guided IOM recommendations about voluntariness informed consent, and confidentiality (Gillon, 2003) In particular the IOM emphasized the necessity for a properly informed public and clientele to minimize harm that could come from the knowledge psychological, familial, or related to insurance or employment discrimination The IOM stressed that informed consent be an essential element of all screening in any setting (IOM, 1994), and Gillon (2003) similarly has argued that respect for autonomy should be "first among equals ' when it comes to adhering to the underlying principles in biomedical ethics

This New Genetics (which encompasses genetic engineering, the mapping and sequencing of the human genome) and the Pandora's box it opens in terms of the complex personal political economic and societal issues will lead to innovative approaches to health and medicine, ethical and legal issues regarding family and economic involvement. The new genetic technologies are rapidly developing and provide unprecedented and expanded opportunities for psychologists to employ their training. The American Psychological Association's (APA's) Ethical Principles of Psychologists and Code of Conduct (APA, 2002) encourages psychologists to aspire to the highest possible standard of ethical conduct for the goal of protecting and promoting the welfare of the people with whom they work. These aspirations include maintaining respect for people's rights and dignity, concern for others' welfare, integrity, competence, and social responsibility. As genetic testing moves quickly from the research realm to the clinical domain and the availability of genetic testing surpasses the number of available genetics counselors, psychologists may be required to take on important new roles and rise to meet the aspirations outlined by their ethics code in the world of the New Genetics.

CHAPTER 2
REVIEW OF THE LITERATURE

The genetic testing/screening literature includes diverse areas such as prenatal screening, carrier screening, predictive genetic testing for late-onset conditions, and newborn screening. For those diseases with a genetic basis, longitudinal studies of those at-risk document the natural history of disease development and progression. Such studies provide the scientific basis for subsequent studies aimed at disease prevention. Providing individuals with personal genetic information also has the potential to facilitate informed decision-making about reproduction and personal risk factor modification. Psychological expertise is needed to aid in delineating the psychological, social, cultural, environmental, and other factors that contribute to the development or prevention of disease and to assist with answering many of the questions regarding the psychosocial, legal, and ethical impact of this technology, including the best methods for communicating complex medical and genetic information, assisting patients and family members with assessing and understanding risks, facilitating informed decision-making, providing psychological support and information about obtaining resources, and evaluating relevant outcomes.

The literature of the risks and benefits of testing is in the formative stage, and is largely descriptive with a few controlled studies to help assess why people engage in genetic testing and how best to present genetic risk information for the purpose of maximizing benefits and minimizing risks, psychological or otherwise. A review of what we do know about the communication of genetic risk information follows.

Complexities of Communicating Genetic Risk Information

In genetics, a risk is defined as the degree of association between particular characteristics and a disease within a defined population (Bottorff, Ratner, Johnson, Lovato, & Joab, 1998). For those at risk individuals, what does it mean to be at an "increased risk" for a particular disease? The ability to predict genetic risk depends on numerous factors related to the test used (sensitivity and specificity), the laboratory procedures, and what we may or may not know about how the disease is transmitted genetically and what triggers expression of the gene. "Increased risk" is a difficult concept for most people to grasp, let alone convey it to others. Risk communication, in genetic counseling literature, involves more than simple exchange of numerical or categorical information. The general public knows little about genetics. Only 50% of those in the general population who were randomly surveyed recently even knew that genes are made up of DNA, and only 29% had heard of the Human Genome Project (Melville, 2001).

Factors Influencing Risk Communication

Little research has been devoted to the best method of communicating complex risk information to people, whether risk communication techniques impact decisions people make about genetic testing or other health related behaviors, or the barriers to risk comprehension. Risk communication and risk perception may be influenced by a multitude of factors and processes. Objective factors, such as method of risk presentation, disease parameters, and the provider of the information (Biesecker et al., 1992), and other subjective factors and experiences that may influence risk perception, either directly or though mediating variables such as anxiety have been identified in the literature These personal factors include cultural beliefs/ethnicity (Donovan & Tucker 2000, Duster & Beeson 1997 Hughes et al , 1997, Flynn, Slovic & Mertz, 1994) educational level (Kong, Barnett, Mosteller, & Youtz, 1986) gender (Shiloh & Saxe 1989) motivation for testing (Lippman-Hand & Fraser 1979a), attitude and expectations, values (Furr & Seger 1998) family or personal history of the disease (Farmer Levy, Turner 1999, Lerman et al , 1994, Rimer 1999) and other psychological variables such as locus of control, coping (Baum et al , 1997) and preference for information (Ader et al , 1992)

In general studies suggest that communication of genetic risk and its interpretation by the at risk individual typically result in a less than ideal understanding of genetic information and decreased retention of accurate information across time (Chase et al , 1983 Gordon Walpole, Zubrick & Bower, 2003, Kessler, 1989), limiting the ability of the at-risk person to make informed choices about the next step to take based on the genetic information provided

Risk presentation. Risk is often communicated using a categorical modifier such as 'low risk" or "high risk " Research examining common verbal expressions of probability has consistently found great variability and ambiguity in qualitative probability words both among laypeople and physicians (Kenney 1981 Hakel 1968) For example, Kenney (1981) had 24 physicians quantify 24 terms and for 15 of these, the response ranged more than 25 percentage points Terms located near the extremes (i e never always) demonstrated less variability Physicians in another study were found to assign values ranging from 1-80% for the word "sometimes" (Bryant & Norman, 1980)

Similar results were reported by Nako and Axelrod (1983), who examined the precision of adjectives and adverbs used to express frequency in the medical literature The majority of qualitative words were found to vary by more than 25 percentage points in probability meaning between physicians and non physicians, making them "too variable for the speaker or writer to be sure that his intended meaning is being communicated when he uses them" (Nako & Axelrod, 1983)

Only one study published in English has examined how *parents* have interpreted qualitative expressions of probability Shaw and Dear (1990) examined mothers understanding of eight common expressions of probability (e g., probably, likely, occasionally unlikely rarely, sometimes) and compared them to physicians' understanding Numerical ratings for seven of the eight expressions differed significantly between mothers and physicians Mothers generally reported a very wide degree of interpretation (e g., on a scale from 0-10, mothers rated "rarely" with numerical probability ranging from 0-8) compared to physicians (physicians rated "rarely" ranging from 0-2.5). Educational level appeared to influence interpretation for three of the variables (rarely, likely, unlikely), in that mothers who left school before the age of 16 differed in their interpretation of these variables from the physicians, while more educated mothers demonstrated no difference. Fifty-three percent of the mothers stated a preference for information in numerical terms, 37% preferred qualitative expressions of probability, and 10% stated no preference (Shaw &Dear, 1990, p.522). Similarly 73% of a sample of women at risk for breast cancer expressed a preference for risk to be described in quantitative terms (Hollowell Stratham, Murton, Green, & Richards 1997) Given that there is a high degree of imprecision associated with qualitative terminology,

---

even among health professionals, many authors suggest that qualitative expressions of frequency be eliminated from medical communications entirely; if qualitative terms are used, then a quantitative value or range should be included (Kenney, 1981; Kong et al., 1986; Nako & Axelrod, 1983; Shaw & Dear, 1990).

Framing effects. Risk perception also appears to be influenced by *framing effects*. Shiloh and Sagi (1989) investigated the meaning attached to quantitative estimates of risk compared to qualitative indicators in a group of undergraduate and graduate students. The authors reported that individuals tend to assign a higher risk when translating from verbal (qualitative) probabilities to numerical (quantitative) probabilities than going from quantitative to qualitative estimates. For example, participants indicated that they thought a 62% probability was "high risk." However, when asked to translate from numbers into words, respondents assigned the category "high risk" a 25% chance (Shiloh & Sagi, 1989). The use of differing denominators can also lead to framing effects. For example, Chase et al. (1983) found pregnant women undergoing prenatal testing were less likely to correctly identify risk equivalents when the denominator was 1000 vs. 100. To illustrate, if one were told that his/her risk of developing a particular disease was 1.3 out of 10,000 compared to the general population risk of 1.0 in 10,000, it may not seem like a large increase. However, reframing this as having a 30% greater risk than the general population may likely be viewed very differently, even though the actual chance of the particular occurrence is nearly equivalent (Slovic, Fischhoff, &Lichtenstein, 1982). Gurm and Litaker (2000) reported that framing an angioplasty procedure as 99% safe resulted in significant differences in hypothetical chest pain treatment participation compared to framing the angioplasty procedure as having a 1 in 100 chance of complications.

Contextual effects. The meaning of a verbal modifier may also change with context (Mapes, 1979; Pepper & Prytulak, 1974; Shiloh & Sagi, 1989). Mapes (1979) reported that physicians' meaning of "rare" changes with the context (e.g., discussing the side effects of beta blockers vs. antihistamines using an identical scale). Merz et al. (1991) found that physicians expressed probabilities associated with a "low chance of infection" substantially higher than the probabilities expressed by a "low chance of death." Women at risk for breast cancer, which is curable if detected early, have been found to tend to inflate their risks (Evans et al., 1993, Kash, Holland, Halper, & Miller, 1992); those at risk for Huntington's disease, which is a fatal progressive disease, have been reported to minimize their risk (Codori & Brandt, 1994).

Cognitive bias. Rita Beck-Black (1982) and Wertz, Sorenson, and Heeron (1986) described several cognitive biases that influence perceptions of risk. Most people tend to organize and assimilate new information according to their existing schemas. An example of this would be an "anchoring bias," or the tendency to bias estimates toward an initial starting value, such that believing one is initially at a high or low risk influences later perceptions of risk, even if evidence is given to the contrary (Nisbett & Ross, 1980). Many individuals who believe they are at high risk for developing Huntington's disease (HD), for example, have difficult time assimilating information that they in fact have a low risk of developing this fatal neuro-degenerative disease. Many of these low-risk individuals remain preoccupied with signs indicating possible disease development (Tibben et al., 1992). The "availability bias" refers to probability estimates based on the ease of recalling occurrences of a particular disorder. Thus, a family history of disease is likely to increase one's risk estimate, as has been demonstrated in a variety of studies (i e , Fanos & Johnson, 1995, Kash et al 1992, Lerman et al , 1993 Tibben et al , 1992) Many people report an "optimistic bias," or a tendency to believe one is less likely to develop a disease than similar peers (Weinstein, 1984 1988) Another is the "gamblers' fallacy," which is the belief that chance does not have a memory (Wexler, 1984) For example it is commonly reported that when couples with a child with a recessive illness such as cystic fibrosis hear their chances of having a child with the disease are one out of four they are often relieved, many believe they have already "used up" their one chance to have a CF child (Fanos & Johnson, 1995) Individuals may also interpret risk in binary terms to simplify the information (it either will or will not happen) (Lippman-Hand & Fraser 1979b, Shilo & Sagi, 1989) Tversky and Kahneman (1981) recommend the use of multiple methods of risk presentation to overcome these cognitive biases involved in risk communication

Methods of Communicating Genetic Risk Information

It has been only recently that the search for the best method of communicating risk has commenced, largely centered on the kindreds with increased breast cancer risks (BRAC1/2 genetic markers) or those at-risk for Huntington's disease (e.g., Bernhardt et al., 1999 Shoda, Mischel, Miller, Diefenbach, Dale, & Engstrom, 1998 Lerman, Schwartz, Miller Daly, Sands, & Rimer 1997, Rimer et al 2001, Sharpe, 1994) Basic research studies of risk perception and randomized trials of alternate risk communication strategies are needed to improve current methods for risk notification (Lerman, 1997) In general the various methods of risk communication involve a genetic counselor and may be done with an at risk family, in person, over the phone in a group setting (Esplen, Toner Hunter, Glendon, Butler & Field 1998) individual counseling with or without an educational component (Kash, Holland, Osborne, & Miller, 1995, Lerman et al., 1995, 1997) using a video (Freda, DeVoore, Valentine-Adams, Bombard, & Merkatz, 1998) and/or using written materials (Browner, Preloran, and Press, 1996, Garrud, Wood, & Stainsby, 2001, Rimer et al., 2001, Wilkinson, Jones, & McBride, 1990). Results of the effectiveness of these communication techniques vary according to demographic (e.g., ethnicity, education, family history) and psychological variables (e.g., anxiety, perceived risk).

For example, Lerman et al. (1995) evaluated the effects of individualized breast cancer risk counseling. Women who were randomized to the counseling group were more likely to improve risk comprehension (subjective risk vs. objective risk) although nearly two-thirds still overestimated their lifetime risk substantially. Those with high levels of anxious preoccupation with breast cancer were less likely to improve, the authors reported that psychological distress appears to be a barrier to comprehension of personal risk information. However, women completing education and counseling were better able to explain the potential risks and benefits of testing for BRCA1. Lerman and colleagues (1997) evaluated the effects of individualized breast cancer risk counseling. Women with first-degree relatives with breast cancer were randomized to education only, education and counseling, and wait-list control conditions. Compared to the wait-list control condition, both risk communication strategies were found to influence knowledge, but not decision to be tested. The counseling approach was superior in increasing knowledge about risks, benefits, and limitations of testing. Increased knowledge of the risks and benefits of testing may be interpreted that the goals of informed consent have been met in allowing the participant to make an informed choice with full knowledge of, and weighting the risks and benefits of, genetic testing.

Much research also indicates that modality of disclosure is important, and that use of videotaped, illustrated and/or simplified language formats improves understanding and the quality of decision making (e.g., Agre, Kurtz, & Krauss, 1994; Jimison, Sher, Appleyard, & LeVernois, 1998). The video format to provide information about serum AFP screening improved comprehension and retention of this information over time and appeared to increase the willingness of the women to participate (Weston, Hannah, & Downes, 1997). Others feel that providers should provide feedback strategies such as recall or recognition tests as part of the process to facilitate understanding (Faden & Beauchamp, 1986). According to these authors, immediate feedback is "simultaneously the best method available for assessing understanding in the context of interpersonal communication and for achieving it" (Faden & Beauchamp, 1986, p. 328).

Finally, providing individuals with written information has also been found to facilitate understanding, decrease anxiety, and increase adherence to behavioral recommendations. A written brochure, which included information on the risks and benefits of hormone replacement therapy, was found to be as effective as a group lecture/discussion format and as an active personalized decision making program in increasing knowledge and satisfaction with the decision-making process (Rothert et al., 1997). Ninety two percent of women attending a genetic counseling clinic for breast/ovarian cancer reported that having a written summary of the genetic consultation served to increase their understanding or recall of information, and 85% intended to use it to assist family members with understanding their risks (Hallowell & Murton, 1998).

Women counseled for familial risk of breast cancer were significantly more likely to retain accurate risk estimates if they received a letter post-counseling reiterating their risk (Evans, Blair, Greenhalgh, Hopwood, & Howell, 1994). Simply sending out a letter regarding the risks of cervical cancer increased the appearance rate for annual exams by 12% (Kash et al.,1992) and including an informational brochure along with a postal notification of an abnormal pap smear result resulted in less anxiety for patients compared to the anxiety of those not receiving the information (Wilkinson, Jones, & McBride, 1990) as well as greater knowledge of a medical procedure, satisfaction with information given, and no increase in anxiety (Garrud et al., 2001). Postcard, letter or phone call reminders have also been reported to be effective in primary care settings in improving immunization rates by 5-20% (Szilagyi et al., 2000). Additional measures such as follow up telephone calls or intervention by medical personnel have been shown to be helpful in facilitating behavior change associated with risk assessment (Champion et al, 2003, Sweet, Willis, Ashida, & Westman, 2003).

Awareness of the factors affecting comprehension needs to be a priority to enhance the quality of the informing process (Edwards, Lilford, Thornton, & Hewison, 1998; Miller and Willner, 1974; Silva and Sorrell, 1988; Tymchuk, 1998; Verheggen, Jonkers, & Kok, 1996). The goal of risk communication should be providing information and clarification regarding the participants' options so that the individual will have the opportunity to make well informed choices. It is surprising that simple, cost-effective strategies, such as providing a written summary and/or reminder letter, have not been used more frequently to increase accurate risk perception, decrease distress, and increase rates of desired screening or other adherence behaviors.

The Impact of Genetic Testing and Risk Communication

Risk communication is important because a person's perception of the risk of developing a disease is believed to be an important determinant of health-related behavior. Perceived risk of developing a disease, rather than actual risk, has frequently been demonstrated to predict interest in genetic testing (Marteau et al., 1991). In addition, perceived risk may influence adherence to screening recommendations through mediating variables such as anxiety (Kash et al., 1992; Lerman et al., 1990, 1993). As mentioned earlier, for those diseases such as diabetes that have a later-onset, genetic testing has the capability to promote lifestyle changes or implement potential treatments to aid in disease prevention. Although relatively few studies have examined the effect at-risk status has on people's health or health behaviors, the need for such knowledge is imperative as genetic testing will continue to become a more powerful tool for disease intervention and prevention.

Accurate communication of risk is particularly important for diseases that may have multiple genetic mutations or that are multi-determined. The test should not cause excessive anxiety but at the same time should not bestow a sense of false reassurance (Axworthy et al., 1996). While a particular mutation might not be found, it does not mean that the individual does not have another mutation not detected by available tests. Similarly, many genes only convey an increased susceptibility (i.e., BRCA1/2, diabetes) and identification of the genes does not imply that one will necessarily get the disease. Not having the "at risk" genes also does not necessarily "protect" a person from developing the disease in question, as there are numerous, not yet understood interactions that come into play when predicting disease risk for multi-factorial diseases. These complexities of the New Genetics highlight the need for a truly informed participant who understands the basic genetic mechanisms benefits and limitations of genetic testing, and implications of results, both positive and negative.

The measurement of the effectiveness of risk communication traditionally includes cognitive outcomes such as knowledge about the test results or accuracy of risk perception or the resultant physical or behavioral changes such as lifestyle modification (Llewellyn-Thomas 1996) Given the fact that numerous factors influence the risk-perception process some (i e , Clarke Parsons & Williams, 1996 Kessler, 1989) argue that recall of risk figures is not the most appropriate indicator of the effectiveness of the risk communication process

Arguments also exist that patient/participant behavior is not an appropriate outcome measure, as in some instances it may imply value statements on personal reproductive decisions (Chapman, 2002), and/or may focus on outcome criteria that professionals perceive to be important (i.e. adherence behaviors) rather than patient/participant-centered concepts such as satisfaction with the process (Edwards & Elwyn, 1999 Kessler 1989, Lippman-Hand & Fraser, 1979a, Michie French, Allanson, Bobrow & Marteau, 1997) In their review of the literature of risk-communication interventions and outcomes, Edwards and Elwyn (1999) report that, in addition to cognitive and behavioral outcomes affective outcomes are becoming increasingly important to measure These include variables such as patient/participant anxiety ratings, satisfaction with the communication process, satisfaction with the information given, assessment of the decision making process or participant certainty that the best choice was made

Cognitive Outcomes and Related Variables

Cognitive outcomes traditionally entail the measurement of participant knowledge post-genetic counseling or some intervention designed to increase knowledge of the disease, the risks and benefits of, or attitudes towards, study participation. Personal risk perception, as calculated by an epidemiological risk, is also commonly assessed either prior to or after risk communication, and accuracy of risk perception assessed. As genetic testing has expanded from prenatal into personal domains, and research methodology and reporting have improved, other literatures have developed, including screening for cancer risk, testing for Huntington's disease risk, and screening for carrier status for diseases such as sickle cell or cystic fibrosis. An examination of variables found to be related to both knowledge and risk perception in these studies will be reviewed, including education, ethnicity, gender, family history, and method of presentation.

Education. Education has been found to be associated with increased accuracy of understanding of risk information in several studies. Parents who had more education were significantly more knowledgeable about newborn screening for cystic fibrosis (Tluczek et al., 1991) and an alfa-feto-protein (AFP) serum test for pregnant women (Browner et al., 1996). This is true not only for genetic studies in particular but also for a broad understanding of information typically used in research, with participants who are less educated unable to define terms commonly used in research such as "protocol" or "efficacy" (Featherstone and Donovan, 1998; Lawson and Adamson, 1995; Taub, Baker & Sturr, 1986; Waggoner & Mayo, 1995). As only 26% of the US population has completed a 4-year college education (National Center for Education Statistics, 2001), it's likely that many genetic research participants will have trouble understanding complex genetic risk information.

Ethnicity. In general, research suggests that differences exist in attitudes, knowledge, and genetic risk perception between ethnic groups, with African American and Caucasian samples most commonly examined. African Americans have been found to report lower levels of knowledge about breast cancer and more positive attitudes about the benefits of genetic testing than Caucasian women (Donovan & Tucker, 2000; Hughes et al., 1997). Data gathered from over 36,000 women attending breast cancer screening programs indicated that African Americans were more likely to perceive their risk to be high compared to Caucasian women (Vernon, Vogel, Halabi, & Bondy, 1993). African American women have been reported to be more likely to benefit from individual breast cancer risk counseling than Caucasian women by significantly improving their personal risk comprehension (60% vs. 22% improvement, Lerman, Lustbader, et al., 1995). Others have also reported that ethnic minority status is related to decreased knowledge about alpha-feto-protein (AFP) screening results (Browner et al., 1996). Caucasian women scored highest after seeing a video and reading a pamphlet, followed by Mexican-American women, and finally recent Mexican immigrants, who traditionally receive little prenatal care. It seems likely that language barriers and differences in cultural attitudes towards genetic information may influence the interpretation of information provided.

Family history. Family history of a disease has been demonstrated to influence risk perception in several studies, both prior to and after risk counseling. For example, it is well documented in the breast cancer screening literature that women who have a family history of breast cancer have elevated and inaccurate perceptions of risk (Braun et

al., 2000, Croyle & Lerman, 1995; Kash et al., 1992, Lerman et al., 1993, Evans, Burnell, Hopwood, & Howell, 1993; Lerman et al., 1995; Lerman, Kash, & Stefanek, 1994, Lerman, Seay, Balshem, & Audrain, 1995; Vernon et al., 1993).

In contrast to elevated perceptions of risk for women with family histories of breast cancer, Farmer, Levy and Turner (1999) reported that a family history of diabetes was associated with increased risk perception in siblings of adult type 2 patients. In contrast, many of those found to be at-risk for Huntington's disease, a fatal neuro-degenerative disorder with no treatment or method of prevention, have been reported to minimize their risk (Codon & Brandt, 1994). Johnson, Riley, Hansen, and Nureck (1990) reported that individuals who were at high risk for developing Type I diabetes tended to minimize their personal risk for disease development, while family members of those at-risk did not engage in minimization of risk status.

Those with no family history of the disease also tend to minimize their risk or report inaccurate risk perceptions or knowledge. Nearly twenty percent of 2900 non-carriers in a cystic fibrosis (CF) community screening program incorrectly believed that there was no risk they were a carrier when in fact they may carry a mutation not detected by the test (Watson et al., 1992). Bekker et al. (1994) found similar results; 17% of those with a negative result believed they were definitely NOT carriers. One third of those with positive results did not understand that they definitely were a carrier.

Method of risk presentation. As previously reviewed in the section on communicating risk in genetics, the method of presentation has been found to influence aspects of genetic education and/or counseling, including the ability to consider risks and benefits (e.g., Lerman et al., 1996) and retention of knowledge across time (Weston et al., 1997) No one method appears to work best for everyone, rather individuals with varying histories and demographics are likely to benefit from a combination of available approaches, some of which are not costly nor time consuming (e.g., use of a video and informational sheet or letter in addition to or instead of traditional genetic counseling) The expanding literature in this area suggests that the most effective intervention is to take a tailored approach for risk communication (Rimer & Glassman, 1999, Rimer et al 2001, Rimer et al 2002)

Time It has been reported that participants are less likely to accurately retain information over a period of time (Chase et al., 1983, Kessler, 1989) For example, the early prenatal genetic counseling literature demonstrates an increase in knowledge of reproductive risk prenatally, although recall and retention of information post-counseling is disappointingly low (see Kessler, 1989 for a review; Ormond, Pergament, & Fine, 1996) Over the long-term, Axworthy et al (1996) found, in a 3-year follow up of those who had undergone population screening for CF carrier status, that 20% of carriers and over 50% of those with a negative results were inaccurate in their recall of their test result, (up from 10% inaccurate at the time of testing for those with negative results) Gordon et al (2003) suggest that inaccurate recall of carrier status in the CF population may be due to memory loss, simplification of the result, and/or risk minimization

Satisfaction with information received Personal preferences come into play too one study reported that participants who believed the amount of information they had received was "just right" had better recall about information found on an informed consent form than those who stated they felt they had received either too little or too much information (Cassileth, Zupkis, Sutton-Smith, & March., 1980) Satisfaction and its

---

relationship to genetic counseling have rarely been assessed, although it is an outcome variable that should be measured more frequently, in order to assist with the development of more patient-centered counseling protocols (Edwards & Elwyn, 1999, Garrud et al., 2003).

In sum, accuracy of information received in genetic counseling studies has been shown to be influenced by many variables, including education, minority status, family history of the disease, and type of communication method. In general, better understanding of risk information occurs with individuals who are more highly educated, non minority members, and who receive individualized/tailored risk counseling, watched a video, and/or receive written follow-up information. However, effects can be variable, tend to diminish over time, and may interact with each other.

Affective Response to Genetic Risk Communication

Accurate risk perceptions appear to be essential to ensure that decisions surrounding genetic testing are made in an informed manner. However, it has been suggested that whether accuracy needs to be increased depends on the consequences of inaccurate recall. For example, women found to be at increased risk for breast cancer may demonstrate discrepancies between their risk status and their behavioral adherence to screening recommendations, such as mammography, even though they may demonstrate accurate recall (Kash et al., 1992). It has been suggested that since accurate recall of risk information does not necessarily lead to risk reducing behavior, one cannot assume that inaccurate recall will impede risk-reducing behaviors (Leventhal, 1989). Therefore, many have begun to examine the links between risk perception and risk-reducing behavior, particularly the potentially mediating variable of disease-specific worry or anxiety

Affective outcomes in the risk communication literature have been gaining ground consistent with a medical philosophy that is more participant-centered than paternalistic (Faden & Beauchamp, 1986). Ratings of affective outcomes might include anxiety, depression, and satisfaction with decision, satisfaction with information obtained, decisional conflict, preference for information, and assessment of information given. Variables affecting only participant anxiety will be briefly reviewed below, including education, family history, optimism, information-seeking style, and time.

Education. There appears to be, for some individuals at risk for breast cancer, an influence of education. Breast cancer risk counseling reduced cancer-specific worry more for those women with lower levels of education (Lerman, Lustbader, Rimer, Daly, Miller, Sands, & Balshem, 1995).

Family history. The effect of family history of disease on reactions to genetic screening protocols is difficult to assess, as most of the research has been studies of groups at risk of disease due to their family history (e.g., BRCA1, HD). In general, notification of at-risk status typically results in an increase in anxiety for carrier screening or testing for diabetes risk (e.g., Johnson & Tercyak, 1995, Watson et al., 1992, Zeesman et al., 1984). For diseases such as Huntington's disease (HD) or breast cancer risk, anxiety or depressive symptoms may last for up to one-year post testing (Bloch, Adam, Wiggins, Huggins, & Hayden, 1992, van't Spijker & ten Kroode, 1997), while others minimize the effect of a positive test results (Tibben et al., 1993b). Besides anxiety, testing for HD has different affective connotations than some other genetic tests. The person at risk either definitely will or will not develop the disease at some unknown point in time. For example, if the person tested does not have the HD gene, and his or her

---

siblings were found to have it, a bond of "at-risk" status could be broken and "survivor guilt" may be instilled, drastically altering family dynamics and relationships, despite supposedly providing relief that one will not develop a neurodegenerative disorder (Codori & Brandt, 1994, Tibben et al., 1992; Sobel & Cowan, 2000). The relationship of family history and anxiety upon risk notification will be an excellent area to continue to examine as screening protocols move from a family based kindred into the general population, who may not have ever experienced or heard of BRACA1 or Huntington's disease.

Dispositional optimism. Scheier and Carver (1985) posit that optimism has important consequences for the way individuals adapt to stressful situations. The tendency to believe that one will experience generally good (as opposed to bad) life outcomes irrespective of one's personal control over those outcomes has been labeled dispositional optimism (Scheier & Carver, 1985). Briefly, dispositional optimism has been found to be related to protection from distress, with higher levels of optimism predicting less anxiety in couples with a failed in-vitro fertilization attempt (Litt, Tennen, Affleck, & Klock, 1992), women undergoing a breast cancer biopsy (Friedman, Nelson, Baer, Lane, Smith, & Dworkin, 1992), men at-risk for developing AIDS (Taylor, Kemeny, Aspinwall, Schneider, Rodriguez, & Herbert, 1992), and women with high risk pregnancies (Lobel, Yali, & Zhu, 1998). Interestingly, dispositional optimism has also been found to be associated with enhanced immune functioning (Cohen et al., 1999, Segerstrom, Taylor, Kemeny, & Fahey, 1998). The mechanism by which optimism may influence distress is through coping (Carver, Pozo, Harris et al., 1993). Optimists have been found to cope differently, and more effectively, with stressors than people who are less optimistic (Scheier, Weintraub, & Carver, 1986, Carver, Scheier, & Weintraub, 1989). In the genetic screening literature, optimism is a new variable for examination. Those individuals low in optimism have been found to be more likely to not engage in genetic testing (Biesecker et al., 2000; Bosompra et al., 2000), consistent with the idea that optimism may provide a protective effect from distress, likely through more effective personal coping strategies.

Information seeking. Miller (1983) hypothesized that individuals cope with health threats in one of two ways, either by seeking more information (monitoring) or by avoiding and distracting oneself from the situation (blunting). In general, an interaction has been reported to exist between the amount of information provided and the monitoring/blunting informational coping style/dispositional trait, matching the amount of information desired and the amount of information received has been shown to lower distress (Ludwick-Rosenthal & Neufeld, 1993, Miller, 1980, Miller and Mangan, 1983). However, whether or not the event is controllable also appears to interact with information given and information preferences to influence anxiety (Miller, Combs, and Stoddard, 1989). Specifically, monitors in genetic testing situations have demonstrated higher distress than blunters. This effect is hypothesized to be mediated by higher perceived risk and more intrusive thoughts in monitors (Schwartz, Lerman, Miller, Daly, & Masny, 1995).

Time. Individuals found to be at-risk for a disease who experience anxiety as a result of learning their risk generally report a reduction in anxiety over time. For example, those individuals found to be CF carriers as adults exhibited a slight increase in anxiety after notification (Clauson, Brandt, Schwartz, & Skovby, 1995), and anxiety from carrier screening programs has been found to subside over a short period (three months) and with proper counseling (Bekker, Denniss, Modell, Bobrow, & Marteau, 1994; Watson, Mayall, Lamb, Chapple, & Williamson, 1992). Johnson and Tercyak (1995) also report that for adult and children at risk for Type I diabetes, a significant increase in anxiety occurs post notification, which dissipates to normal levels over a period of about three months.

To sum, affective variables are increasingly important to measure. Many variables have been shown to influence anxiety. For example, some interventions appear to be more effective for those with lower levels of education, and the response depends on the disease for which risk is being communicated. Individuals who tend to have dispositional optimism report being buffered from stress by using effective coping strategies, and information-seekers tend to be more anxious in situations in which they may think more about their perceived risk. While anxiety may be elevated for many individuals told they are at-risk, it has been reported to return to levels within normal limits from 3-12 months post-testing. A consideration of an individual's personal and disease characteristics will help to tailor the counseling so that anxiety is not unnecessarily inflated.

Behavioral Outcomes in Genetic Risk Communication Studies

Behavior as an outcome measure has been used in the risk communication literature for years, generally focusing on adherence to treatments, tests, or trials. It may be measured as intended behavior or actual observed behavior. There has been a shift in emphasis from "compliance" with medical prescription to a more participant-centered approach that assesses adherence in light of what the participant wants, such as taking hormone replacement therapy or not (Rothert et al., 1997), or participating in genetic

---

screening protocols or not Variables affecting behavioral outcomes that will be examined include gender ethnicity education altruistic motives information-seeking perceived risk and disease-specific anxiety

Gender Females have been found in several studies to be more likely to participate in predictive genetic testing studies for Huntington's disease (Holloway et al , 1994, Meissen et al., 1988 Tibben et al , 1992) diabetes (Farmer et al , 1999), and breast cancer (Lerman et al , 1996) Women typically act as the "genetic housekeeper" (Richards 1996) often initiating contact with the testing center and gathering information.

Ethnicity. A recent study by Tucker and Pedersen (2000) examined ethnic differences in health care preferences and found that Hispanics preferred a doctor who could speak Spanish, and African Americans reported a mis trust of Caucasian physicians and fearfulness that they would be used as "guinea pigs." African Americans in particular have reported a bias toward not participating in research due to the injustices suffered by the African American community prior to regulation of research (e.g., Tuskegee syphilis study (Jones, 1981 Howard-Jones, 1982), and the discrimination caused by inadequate public education about carrier status for sickle cell screening programs (Hampton, Anderson, Lavizzo, & Bergman, 1974)

Education. Harth and colleagues (1992) examined parents who had volunteered their children for clinical research versus those who had not and found a "psychosocial filter" effect, those parents who volunteered were less educated and had lower SES (Harth, Johnstone & Thong, 1992, Harth & Thong, 1991, Silverman, 1989) Similarly, among parents responding to a hypothetical situation regarding the enrollment of their healthy newborns in a clinical trial, those who indicated refusal (74%) tended to be more educated and more concerned about the risks of research and possible inefficacy (Duterte, Barbier, Jonville, Pierre, & Berger, 1993). However, opposite results are seen for those obtaining personal genetic information. For example, women requesting genetic results for breast cancer risk had higher levels of education and perceived the benefits of testing to be important than those not requesting results (Lerman et al., 1996, 1999). Those accepting testing for Huntington's disease also had higher levels of education than the average person (Codori, Slavney, Young et al., 1997).

Altruism. Oliner and Oliner (1988) define a behavior as altruistic when it involves a risk, no external reward, is directed toward helping another, and is voluntary. Altruism as a motive for participating in studies which may provide no personal benefit should be examined. However, the research on measurement of altruism is sadly lacking (Rushton, Chrisjohn, & Fekken, 1981) and most measure altruistic participation anecdotally. Why would parents have their child participate in potentially risky research studies that may or may not have any direct benefit to them? Over half of 181 parents/guardians who had enrolled their child in a randomized, double blind, placebo controlled trial of ibuprofen for the prevention of febrile seizures stated they participated to further the goals of science (van Stuijvenberg et al., 1998). However, study participants who stated they participated out of altruistic reasons, also believed that through participation in the study they might be able to gain better access to care or other personal benefit (Childs, Gordis, Kaback, & Kazazian, 1976, Green, Murton, & Statham, 1993, Harth & Thong, 1991).

Information-seeking Monitors, or those who seek information, have been shown to exhibit better adherence to behavioral recommendations, such as cancer screening (Wardle Collins, Pernet, Whitehead, Bourne, & Campbell, 1995), although the literature is too limited in this area to make further generalizations

Perceived risk. Those individuals who perceive themselves to be at a high risk for a disease tend show interest in testing (Marteau et al 1992 1991) Codori et al (1999) reported that among those who had a high-degree of worry/thoughts about getting hereditary non polyposis colorectal cancer, the probability of genetic testing increased as perceived risk increased Among those who rarely thought about getting colorectal cancer, risk perception was unrelated to the testing decision The general research in this area suggests that while perceived risk may influence uptake of testing, it appears that risk perception likely influences the decision to be tested through the mediating variable of disease-specific anxiety

Disease-specific anxiety Many studies report that a large majority of both members of the general population and at-risk groups express favorable attitudes toward and interest in being tested for diseases such as HD, colon cancer, breast cancer, neonatal screening, and carrier screening to name a few However, high rates of interest do not necessarily lead to high rates of utilization, and several researchers have questioned the discrepancies between attitudes and behavior that have been reported in this literature (Markel et al 1987, Meissen & Berchek, 1987, Quaid et al , 1989) It is likely that anxiety or disease specific worry plays a role in the decision to be tested or not.

For example Lerman Rimer et al (1994) found that 95% of those at-risk for breast cancer reported they would or probably would use testing for BRCA1 Cancer-specific worry has been shown to be related to intentions to use genetic testing (Cameron & Diefenbach, 2001, Croyle & Lerman, 1995, Kash, Ortega Verdejo, Dabney Holland, Miller, & Osbourne 2000, Lerman, Daly, Masny, & Balshem, 1994) cancer-specific worry in at-risk families has been reported to range from 43%-80% (Botkin Croyle et al , 1996 Lerman et al, 1996) Those more likely to request results of genetic testing had a high degree of cancer-specific worry (Lerman et al ,1996) and after controlling for demographic variables, cancer-specific distress predicted use of BRCA1 screening (Lerman, Schwartz, Narod Lin, Hughes & Luyneh, 1997) Worry was also found to be the most important variable influencing decision about prenatal testing in a group of at-risk women (Kenen, Smith Watkins & Zuber Pittore, 2000)

On the one hand the perception that one is at-risk can lead to increased vigilance and protective behavior to reduce a health threat On the other hand, extreme levels of anxiety can interfere with adherence behavior (Janis & Feshbach, 1953) Prior to the availability of genetic testing, anxiety was found to be a deterrent to screening behaviors (i e , mammography pap smear) for women at high risk of cancer (Kash et al , 1992) Lerman and colleagues have found that higher anxiety and cancer worries have been associated with lower rates of mammography (Lerman et al., 1990, 1993), lower rates of breast-self exam (BSE Alagna et al., 1987) and lower rates of clinical breast examination (Kash et al. 1992) Lerman and colleagues also looked at breast screening practices among women at risk due to prior abnormal mammograms and found a curvilinear relationship between breast cancer worries and BSE practice (Lerman et al, 1991) Their findings suggest that women with moderate levels of cancer worry were most likely to practice monthly BSE than women with high or low levels of worry Some degree of anxiety also appears necessary to motivate women with abnormal mammogram results to obtain repeat screening. Lerman and colleagues (1991) reported that 77% of women with abnormal results had obtained repeat mammography within 1 year; those who did not repeat the mammography reported the lowest level of worry. Lerman, Miller et al. (1991) also found that heightened anxiety after abnormal pap smear test results led to avoidance of a confirmatory test, while those who obtained repeat testing had lower levels of worry. Lerman, Schwartz et al. (1997) note, it makes sense that distress promotes risk assessment in the cancer screening literature, as knowing one is at-risk affords you the opportunity to either reduce uncertainty, minimize distress, or provide you with the opportunity to consider methods of prevention rather than simply adhering to disease detection efforts.

As just reviewed, behavioral outcomes, in terms of participation rates in genetic studies, are likely to be affected by gender (females more likely), ethnicity (minorities less likely), education (those who volunteer their children in research tend to have lower levels of education while those seeking personal information tend to be more highly educated). Higher amounts of information-seeking and altruistic feelings may increase likelihood of participation. Perceived risk appears to increase participation by increasing anxiety levels, which may serve to influence behavior depending on the characteristics of the disease being tested. It appears that there may be an optimal level of anxiety necessary to motivate testing, while at the same time not too much anxiety that might lead to avoidance behavior. Examining outcomes like these by utilizing various theoretical models may help one to assess the relevant variables related to the appraisal of risk, affective response, and the resultant behaviors.

Theoretical Models Predicting Effectiveness of Risk Communication Techniques

The majority of studies examining intentions to engage in genetic testing or assessing utilization of the information obtained as a result of genetic testing are purely descriptive in nature. Most fail to use any theoretical model to inform study design or interpret findings. The use of theoretical models to guide and interpret research efforts in the area of genetic screening is needed for the development and evaluation of the outcomes of various risk communication strategies.

Several researchers have used the Health Belief Model (HBM; Rosenstock, 1974) in genetic screening/counseling programs or reported a link between HBM model components and decisions about testing (i.e., Becker et al., 1975; Frets et al., 1990, O'Connor, 1996, Rowley, Loader, Sutera, Walden, and Kozyra, 1991, Sagi, Shiloh, and Cohen, 1992, Shiloh and Saxe, 1989, Sorenson et al., 1987). This model predicts health behavior in terms of the individual perceptions about the threat of disease and the efficacy of treatment in reducing the threat. It states that health surveillance behavior is based on beliefs about the health threat, including: (1) perceived disease susceptibility and (2) perceived disease severity; and beliefs about the costs/benefits of a specific health behavior, including the (3) potential benefits of a health action or behavior in reducing the threat, and (4) barriers to actions (costs, risks, inconveniences). These four beliefs and their interaction with demographic variables, along with cues to action (some internal or external event initiating the health behavior) result in the decision. For example, those who participated in a Tay Sachs screening program were more likely to perceive they were carriers (perceived susceptibility) and less likely to anticipate negative psychological consequences than those who chose not to participate (Becker et al., 1975).

Lerman et al. (1997) suggested the Self-Regulation Model of Health Behavior (Leventhal 1965) may help to explain findings that women at risk for breast cancer who exhibited either too high or too little worry were less likely to practice risk reduction behavior. The Self-Regulation model suggests that moderate levels of perceived health threat (e.g., diagnosis of cancer in a relative) will motivate persons to adopt a health practice in order to reduce the anxiety generated by the health threat. Some individuals with low levels of anxiety may hold an optimistic bias regarding their personal risk and be less likely to adopt preventative health practices (Weinstein 1988). While excessive cancer related anxiety might produce avoidance of screening, at least minimal levels of anxiety may be necessary to motivate these behaviors.

The Fear Arousing Communications Theory (Janis & Feshbach, 1953) also states that a moderate level of arousal of fear is considered optimal for producing the engagement in preventative health care behaviors. Too little arousal may promote denial of risk and too much arousal may lead to avoidance. The nature of the behavioral outcome (i.e., opportunity for prevention or detection of a hard-to treat or even incurable disease) also has implications for the use of this model (Lerman, Schwartz et al., 1997 Sweet et al., 2003).

Baum, Friedman and Zakowski (1997) have discussed the use of a rather comprehensive model of stress and genetic testing for disease risk. Based on Lazarus & Folkman's (1984) transactional model of stress and coping, the Stress-Disease Risk Coping Model was proposed to explain the interaction between personal variables (i.e., appraisal of risk, coping resources) and the stressor characteristics (i.e., nature of result, disease characteristics, uncertainty reduction) involved in genetic testing. Specifically they hypothesize that based on this model, distress will be affected by the interaction between personal factors (perceived risk influenced by family history, optimism), the actual test results, characteristics of disease, and degree of uncertainty remaining after testing. This model also identifies some of the behavioral consequences of distress that in turn may influence health and disease. For example, too much or too little anxiety may lead to reduced surveillance behaviors, as previously proposed by the Fear Arousing Communications Theory. Baum et al. (1997) propose that a comprehensive model of stress and coping, including assessment of mediating or moderating personal variables, be used to aid in examining the effects of evaluations for genetic risk. The present study will address several of these variables.

Newborn Genetic Screening

Making the decision to undergo genetic testing is often complicated enough, but the complexity increases when children are the potential subjects. Several groups now have guidelines regarding genetic testing of children (American Academy of Pediatrics, 2001, American Society of Human Genetics/American College of Medical Genetics, Boards of Directors, 1995). Important factors to consider when testing children for genetic disorders, particular if there is no known cure, include the psychological, social, and economic effects of testing, impact on the family dynamics, impact on future planning, remoteness of time of disease onset, degree of risk, possible future medical benefits, level of maturity of the child, and ethical principles, such as respect for persons and beneficence.

Much controversy surrounds predictive genetic screening for chronic and life-threatening diseases with no-known cures. Modell (1992) describes three criteria for genetic screening to be beneficial 1) a clear diagnosis 2) the possibility of providing clear information on risk and 3) an effective solution Guidelines exist which do not advocate the testing of children unless some immediate medical benefit is present, except in the context of regulated research protocols (IOM, 1994, American Society of Human Genetics/American College of Medical Genetics Board of Directors, 1995 Wertz et al., 1994) Typically, newborn screening is encouraged only if there is evidence that a newborn would benefit from effective treatment identified as early as possible

Benefits and Risks of Newborn Genetic Screening

Newborn screening is the most widely used type of genetic screening, with nearly all states mandating newborn screening for phenylketonuria (PKU) and congenital hypothyroidism Undetected, these diseases can lead to mental retardation but if identified and treated early normal development can be assured. Testing is generally only offered for early detection and risk reduction Some of the drawbacks include the high rate of false positives, up to 50 times greater than those congenital disorders actually detected (Kwon & Farrell, 2000) Screening programs that have high false positive rates have been reported to generate both acute and chronic parental anxiety, along with more negative perceptions of their child's health, in those screened and later found to be negative for diseases including PKU ("The PKU Anxiety Syndrome", Rothenberg & Sills 1968) congenital hypothyroidism (Bodegard, Fyro & Larsson, 1983) and cystic fibrosis (Tluczek et al., 1991)

With diseases such as sickle cell or other hemoglobinopathies not only are affected individuals identified but carriers as well This requires careful attention to the education and counseling of these parents to prevent stigmatization and labeling of the child without adequate knowledge of what the test result means (Hampton, Anderson, Lavizzo & Bergman, 1974, Markel 1992) Hampton et al (1974) initially reported that nearly half of parents of children found to be sickle cell carriers thought of their child has having a disease with two-thirds believing their child needed dietary supplements to maintain health However as screening methods have improved, and genetic tests used have increasing sensitivity and specificity along with improved methods of risk communication or genetic counseling, these potentially adverse effects are likely to be minimized To date the psychological implications of screening newborns have rarely been examined Studies reporting on the effects of newborn screening programs will be briefly reviewed In many studies there have been conflicting findings on the psychological impact of various screening programs for disease detection and risk reduction

A small literature exists on newborn screening for Duchenne muscular-dystrophy, an incurable X linked condition eventually leading to death during early adulthood This screening has been implemented in Wales to reduce the delay in diagnosis (Fenton-May et al 1994) for newborns in at-risk families Of the small number of children screened, all but one family had favorable attitudes towards early diagnosis about their son s condition. One family reported that they had not been provided with the information sheet prior to testing, and would have refused the newborn screening given the significant negative emotional impact of the news on their family (Bowman, 1993, Bradley, Parsons, & Clarke, 1993)

Another interesting cohort includes Swedish newborns in the mid 1990 s that were screened for alpha 1-antitrypsin deficiency a recessive condition resulting in lung disease. Identification of at-risk infants was implemented to provide information regarding the natural history of the disease and the possibility to reduce risk from exposure to air pollutants However the screening program was reportedly discontinued when it was found that, at follow-up, parental smoking in the home had actually increased Fathers even smoked twice as much as fathers of children without the condition. Parents viewed their child as more sickly and with problematic behaviors There did not appear to be any medical benefit to newborn screening and, in fact, this particular screening resulted in "paradoxical adherence" to medical recommendations (McNeil, Sveger & Thelin 1988) meaning parents with at risk children advised to smoke less actually smoked more than those whose children were not at-risk. In addition, concepts such as "pre selection" of a child to have disease or a "vulnerable child" and subsequent symptom searching (Tibben et al , 1992), have been reported in the literature in those families at-risk for genetic disorders such as Huntington s disease (Kessler, 1988), for which newborn screening is not advocated (AAP, 2001)

Genetic screening for cystic fibrosis (CF, Kerem et al., 1989) has received the most attention in the newborn genetic screening literature. Screening programs exist that are both population based as well as limited to those with a family history of CF For example 92 % of Australian-born children are tested, while only 6% of USA newborns are screened, most in Wisconsin or Colorado where it is state-mandated (Wilken & Travert, 1999) With no clear consensus as to medical and psychological effects of screening, many have expressed concern regarding the potential psychological effect of early screening Some argue that early diagnosis provides no benefit and that knowledge without effective treatment may be psychologically harmful especially for those with no family history due to the unexpected results (Wald & Morris, 1998). Some mothers have reported a temporary rejection of their child either during the testing or diagnosis of CF via early screening (Al-Jader, Goodchild, & Harper, 1990) and some families have reported significantly elevated parenting stress and a greater likelihood of viewing their child as demanding (Baroni, Anderson, & Mischler, 1999). However, delay in diagnosis when newborn screening was not done has been associated with increased maternal distress (Al-Jader et al., 1990; Boland & Thompson, 1990; Wilken, Towns, & Mellis, 1983). In addition, early diagnosis is viewed by many as an opportunity to provide early treatment, with increased nutritional outcomes (Farrell et al., 1997), improved lung functioning (Walters, Wilken, Irwig, Simpson, Van Asperen, & Gaskin, 1999), opportunities for research for effective treatments, and parental awareness that they are carriers of CF for use in reproductive planning (Lane, Harris & Harris, 1998).

Screening programs for young children at-risk of a childhood-onset disorder, such as familial adenomatous polyposis (FAP), have been reported to reduce parental distress due to a reduction in uncertainty after testing (Marteau & Michie, 1995), by providing the opportunity for preventative action, allowing for benefits such as time to adjust psychologically to risk status, and/or relief from invasive yearly cancer screening. Similarly, children at risk for multiple endocrine neoplasm type 2 routinely undergo genetic testing, as prophylactic thyroidectomy prior to developing certain cancer has reduced mortality in these children (Wells & Skinner, 1998). In these cases, opportunities exist for disease prevention or surveillance options, the conclusions we can draw for screening for diseases that as of yet have no effective method of prevention are limited

---

Documenting the psychological effects of newborn screening protocols is a new area of research and will be crucial to examine the family's understanding of the screening, responses to testing, and whether or not the screening may lead to positive health or other psychological or behavioral outcomes. It is unclear yet whether population based screening programs have a higher potential for psychosocial harm than screening programs for at-risk families. Adequate information provision or genetic counseling will be crucial to reduce distress and increase satisfaction and good decision-making regardless of a personal or familial risk for disease.

The Prediction and Prevention of Type 1 Diabetes

The prevalence of Type 1 diabetes is greater than that of all other childhood diseases combined and is on the rise (LaPorte, Matsushima, & Chang, 1999). Models of disease development indicate that a combination of genetic and environmental factors appear to be involved. For example, the concordance rate for Type 1 development in identical twins is between 20-50%, with a 2-8% rate for siblings or parents (Kyvik et al., 1995; LaPort et al., 1999). However, nearly 90% of Type 1 diabetes occurs in families with no history of the disease (Dahlquist, Blom, Holmgren, Hogglof, Larsson, & Sterky, 1985).

Researchers have identified the HLA gene region of the short arm of chromosome 6 (the Major Histocompatability Complex (MHC) as having a 40-60% correlation with disease development (Davis et al., 1994; Krip, 1998). The particular alleles of HLA-DR3/4 and HLA-DQ have been shown to confer an especially high risk of disease development (refer to Appendix A for a table of risk relative to genetic make-up). The presence of humoral immune markers, such as islet cell antigens (ICA) (Bruining et al.,

---

1989, Riley et al , 1990, Schatz et al , 1994), autoantibodies to insulin (IAA) (Landin Olsson et al , 1992, Roll Christie, Standl, & Ziegler, 1994) GAD (Baekkeskov et al 1987), or islet antigen-2 (IA-2) have been found to greatly increase one's risk of disease development. A direct relationship has been established between the number of detectable autoantibodies particularly ICAs, and risk for Type 1 diabetes (Knip, 1998) These markers may be present years before disease onset (Riley et al , 1990) Thus identification of at-risk individuals provides a unique opportunity to study the natural history of disease for the purposes of disease prediction, intervention, and ultimately prevention

Little is known about the psychological impact of participating in prediction and prevention trials for Type 1 diabetes Currently trials are underway in which newborns at-risk for Type 1 diabetes will be followed over the course of many years to elucidate the factors contributing to the pathogenesis of disease (e g , Akerblom 1993 Rewers et al 1996 Schatz et al , 2000, Wagner et al. 1996 Ziegler Hummel Schenker & Bonifacio, 1999) It is necessary to study genetically predisposed and genetically protected infants to increase our understanding of the pathogenesis of diabetes and to carve pathways for prevention efforts In the past, the study of the natural history of diabetes has typically been conducted with older children and adults and requires invasive and long term monitoring of at-risk persons As genetic screening protocols move from small family-based protocols into large, population-based screening efforts, it will be important for the establishment and maintenance of these at-risk registries to document the individual and familial understanding of, and reactions to the news of risk and to develop appropriate educational and counseling interventions (Tercyak, Streisand, Peshkin, & Lerman 2000)

Previous research conducted in our laboratory has documented significant psychological sequelae associated with the use of screening programs to identify persons at risk for the development of Type 1 diabetes in both adults and children (Johnson, 1997, Johnson, Riley, Hansen, & Nurick, 1990, Johnson & Tercyak, 1995, Johnson & Carmichael, 2000). In general, at-risk notification has been found to induce clinically significant anxiety in the at risk individual and in family members, especially mothers of at-risk children (Johnson, Baughcum, Carmichael, She, & Schatz (in press), Johnson & Tercyak, 1995, Johnson & Carmichael, 2000). Specifically, Johnson and Tercyak (1995) examined the influence of positive islet cell antibody (ICA+) screening results on anxiety levels, as measured by the state portion of the State-Trait Anxiety Inventory (STAI, Spielberger, 1983) and the State-Trait Anxiety Inventory for Children (STAI-C, Spielberger, 1973). Those testing ICA+ had approximately a 50% chance of developing diabetes within the next five years. Results indicated that ICA+ children, ICA+ adults, and their family members experienced clinically and statistically significant levels of anxiety upon notification of ICA+ status. After four months, anxiety had returned to normal levels. Reported anxiety was higher for parents of ICA+ children than it was for spouses of ICA+ adults. In addition, parent and child anxiety was significantly positively correlated. Greater levels of anxiety were associated with lifestyle or behavior modification. Fifty-two percent of ICA+ children and 24% of ICA+ adults reported altering their behaviors and/or lifestyle in an attempt to delay or prevent the onset of Type 1 diabetes.

Preliminary work in a newborn screening population. Little research has examined parents' psychological reactions to participation in a newborn screening program for Type 1 diabetes, although parents have indicated favorable attitudes towards risk screening and prevention trials for Type 1 diabetes (Lucidarme, Domingues Murial, Castro, Czernichow, & Levy-Marchal, 1998; Tercyak, Johnson, and Schatz, 1998). Yu et al. (1999) used the total stress score (TSS) of the Parenting Stress Index (PSI, Abidin 1990) to assess maternal parenting stress levels 5-7 weeks after delivery of infants and again 4-5 months after the mothers had been notified of their newborns' genetic risk status. There was no relationship between risk level and change in TSS. However, the median decrease of TSS of mothers of infants at high risk was smaller than the median change for mothers of low risk newborns. A limitation of this study is that the PSI was intended to measure general parenting stress, not stress specific to diabetes-risk. Also, the PSI may not be an appropriate measure for parenting stress in infants.

In our laboratory, Carmichael, Johnson, Baughcum, Fuller, Taylor, She, and Schatz (2003) and Johnson, Baughcum, Carmichael, She, and Schatz (in press) interviewed 351 mothers of infants identified at birth as at-risk for developing type 1 diabetes (through the presence of HLA-DR and HLA-DQB1 alleles) in the longitudinal Prospective Assessment of Newborns for Diabetes Autoimmunity (PANDA) study. Participation rate was high; for the first Follow-Up Interview, 91% (N=351) of the mothers contacted (of over 600 eligible) agreed to be interviewed initially, and 78% consented to participate in a Follow-Up Interview 2 (N=232).

These mothers were largely Caucasian (75.6%), with 12.1% Hispanic, 6.8% African American, and 1.8% Asian. The majority of mothers were married (76.2%) and highly educated: 69.6% reported having some college education, with a median income in the $30,000-$40,000 range. In this sample, 68.1% of the mothers initially stated they wanted to participate in the longitudinal study when the study nurse informed them of the baby's at-risk status; 6.2% immediately declined, and the remainder of mothers reported indecision about their infant's future study participation. The majority of the sample was identified as at "moderate" risk (63.5%), with 32.8% at "high" risk and 3.2% at "extremely high" risk. Over one quarter (26.2%) of the sample reported no known family history of diabetes of any type. Eight percent claimed to have a first-degree relative with type I diabetes, and the remainder reported having a second-degree (or greater) relative with diabetes (any type).

Our assessments were linked with standard procedures in this longitudinal newborn genetic screening study implemented at the University of Florida. Infants in the PANDA study were screened at birth for the presence of HLA-DR and HLA-DQB1 alleles using the Guthrie test (obtained by heel stick at the time of the state-mandated PKU testing). Those parents with infants at an increased risk (i.e., moderate, high, extremely high) were contacted by phone approximately one to two months after the birth by the study nurse. Using a standardized script, mothers of babies identified as moderate, high, or extremely high risk were informed of their child's results, using both categorical terminology (i.e., moderate, high, extremely high) and numerical estimates (2%-25%) of risk, as suggested by the literature.

At the time of the notification call, the nurse also asked the parents to consider continued participation with the study, which involved further blood draws at 3, 6, or 12 month intervals to monitor the development of autoantibodies and other biological markers associated with the development of type I diabetes. Mothers were also asked for permission for our research team to contact them for a phone interview. Those mothers who agreed to participate in our study were interviewed by phone approximately 1–2 weeks (Follow-Up Interview 1) and again 2-4 months (Follow Up Interview 2) after notification of at-risk status. All mothers of at-risk infants (including those who were undecided or refused the longitudinal FANDA study) were subsequently called by our laboratory staff to request participation in our interview.

Maternal accuracy of risk perception. As indicated previously, the study design included a standardized "scripted" presentation of neonatal risk status by the study nurse using both a descriptive label (moderate risk, high risk, extremely high risk) and a numerical estimate of risk. This allows us to compare mothers' reports of the baby's risk status with what the mother was actually told using the scripted protocol. At Follow Up Interview 1 (conducted within one-two weeks after the mother was first informed of the baby's at risk status by the study nurse), nearly 32% of mothers incorrectly recalled their child's risk information: 4.3% overestimated their child's risk, 14% underestimated their child's risk, and 13.8% could not recall any risk at all, even after given six categorical and numerical risk estimates for recognition purposes. Approximately 2 months later we conducted Follow-Up Interview 2 and again assessed maternal understanding of the infant's risk status. Over time, more mothers minimized their child's risk, with 23.7% underestimating their child's risk at Follow-Up Interview 2 (compared to 14% underestimating the risk at Follow Up Interview 1). Fewer (2.6%) mothers continued to overestimate the baby's risk (compared to 4.3% at Follow-Up Interview 1), 58.6% were accurate about the baby's risk status (a decline from 68% at Follow-Up Interview 1), and 15.1% could not remember the risk (14% could not remember at Follow-Up Interview 1)

An analysis of the responses of mothers when questioned about their child's risk, both spontaneous and prompted, showed that nearly 39% of mom's re-stated their child's risk in categorical terms, whether accurate or not (i.e., low, moderate, or high risk). Nearly 19% cited a numerical risk estimate. Thirty-one percent of mothers reported recalling both a category and a number for their child's risk, while ten percent were unable to recall a category or a number.

Mothers were also asked whether they believed their child would ever develop diabetes. Initially, the majority (75.3%) indicated that they were unsure about their child's ultimate health status, although 17.5% stated that their child would never develop diabetes and nearly 7% indicated that they expected the child to ultimately become diabetic. At Follow-Up Interview 2, fewer mothers (65.4%) remained unsure of their baby's health status. Some (10%) were convinced the child would ultimately develop diabetes. However the largest shift occurred in the number of mothers who believed the child would *never* develop the disease: 25.5% (compared to 17.5% at Follow-Up Interview 1).

Logistic regression analyses were used to assess maternal and infant variables that may be related to maternal accuracy about the baby's risk status. At the time of Follow-Up Interview 1, mothers having lower levels of education ($B=-.380$, $p<.001$, $R=-.151$), and those from an ethnic minority group ($B=-.78$, $<.005$, $R=-.12$), were less likely to be accurate about their child's risk. Initial risk classification, family history or initial anxiety was unrelated to accuracy of risk perception. Upon examination of predictors of maternal accuracy about the baby's risk status at the time of Follow-Up Interview 2, approximately 2.4 months later and immediately before the baby's first scheduled follow up blood draw, maternal accuracy was best predicted by initial accuracy of risk perception ($B=2.34$ <000 $R=-42$) and family history of diabetes ($B=40$, $<.023$, $R=11$) Mothers who were accurate about their baby's risk status at Follow-Up Interview I tended to remain accurate over time In addition, mothers who reported a family history of diabetes were also more accurate in their recall of risk information

These data suggest that mothers given unfavorable news about their infants health, may tend to underestimate the baby's risk over time and to develop greater confidence that the child will never develop the disease This is particularly true for those who do not have a family history of diabetes of any type and for those who were initially inaccurate in their recall of their child's risk. These findings are consistent with previous findings that individuals given information about diabetes risk tend to minimize at-risk status as a coping strategy (Johnson & Tercyak, 1995). When communicating information about diabetes risk to mothers of at-risk newborns, particular care may be needed with mothers having lower levels of education, no family history of diabetes and mothers from minority groups. This is in line with the literature on screening programs and maternal understanding. Simple but effective interventions to ensure the goals of informed consent are realized and to promote accurate understanding of risk information and informed decision making about study participation are needed.

Maternal anxiety Consistent with previous research we found that maternal anxiety levels were clinically elevated after initial notification of infant risk status and decreased to normal levels after a period of 2.4 months ($t=12.54$ $p<01$) Initial anxiety was significantly positively correlated with anxiety over time ($r=63$) as well as the use of all types of coping styles, both problem and emotion-focused Higher maternal anxiety immediately after at-risk notification was associated with higher infant risk (B=1 53, p<.04), lower maternal education (B=-.907, p<.041), and ethnic minority status (B=-.265, p<.031) Maintenance of higher maternal anxiety over time was associated with higher initial anxiety upon hearing of infant risk (B=.406 p<.00) high use of wishful-thinking (B=9.005, p<.00) a family history of any type of diabetes (B=2.355, p<.002), and use of self blame regardless of initial anxiety level (B=.189, p<.006) This model accounted for approximately 49% of the variance in anxiety at the second assessment, 2 4 months after the first assessment

Continued study participation. Upon initial notification of infant at risk status, 68 1% of 627 mothers who were informed by the study nurse that their child was at moderate or high risk stated an interest in continuing with the study Over a quarter (25.7%) of the mothers indicated they needed time to think about the information before making a decision, and 6.2% immediately reported no interest in further participation in the study Logistic regression indicated that mothers with an infant at a higher risk (B=.49, p<.045) and mothers with higher levels of anxiety at the time of notification (B=-.03 p<.000) were significantly more likely to state a wish to continue in the study initially However, only 36 8% (N=331) of 726 mothers had either scheduled an appointment for the blood draw or had actually brought their child in for the blood draw by the time of data analysis One-third of the mothers (N=205) were classified as "inactive," indicating that while they initially stated a desire to continue with the PANDA study they either did not schedule an appointment (N=111), or they scheduled an appointment and did not appear for it (N=94) Nearly 17% reported they no longer wished to continue up over 10% from their initial statements indicating a loss of interest over time for these mothers. Stating one would indeed like to continue and participate in Part II of PANDA over time (regardless of whether they scheduled or appeared for a blood draw) was associated with a higher infant risk (B= 08, $p< 001$) and having a family history of any type of diabetes (B= 65 $p< 054$)

Among mothers initially stating they would like to continue with PANDA, those who showed for the blood draw versus those who did not show or did not schedule an appointment had babies with a higher risk (B= 528,$p< 004$), higher maternal anxiety (B=-038 < 028), and a family history of diabetes (B=.526 $p< 072$) For those mothers who made the blood draw appointment, the sole predector of appearing for the blood draw was infant risk (B=1 51, $p<.000$) Those mothers with an infant with a higher actual risk were significantly more likely to appear (50 5 %) than those with infants at moderate risk (28.2%) It is unclear whether mothers with infants at a higher risk were targeted more by study staff for continued participation than mothers of infants at a lower risk, and this will need to be examined in future studies

As only ~20% of infants screened are found to be at increased risk, and only a third of those have either scheduled or appeared for the PANDA Part II blood draw (Schatz et al , 2000) an examination of the variables that promote participation in the newborn registry is important. The application of theoretical models and/or the development of effective interventions may help to increase retention rates of at-risk infants from the general population to provide data important to the prediction of Type 1 diabetes

CHAPTER 3
RATIONALE FOR AND PURPOSES OF THE STUDY

For some diseases such as diabetes, genetic risk information only provides part of the story, as disease development is determined by complex factors that require prospective study in genetically susceptible individuals. However, population-based genetic screening programs target individuals who may or may not have previous experience with diabetes. Participation in genetic screening programs may not provide additional health benefits to the individuals screened, although population based screening may be necessary to understand the natural history of the disease and the discovery of a cure.

Making the decision to participate in genetic screening studies is not easy and may be influenced by a variety of factors, including risk perception, education and ethnicity, previous experience with diabetes, information-seeking style, dispositional optimism, and anxiety. One purpose of this study was to evaluate the impact of a brief educational letter sent to the parents of an at risk infant identified by newborn genetic screening. The effect of the educational letter on cognitive, affective, and behavioral outcomes, including perceived risk, diabetes specific worry, and stated intentions to continue participation in the longitudinal diabetes-screening program, was examined. In this population, no intervention to increase accuracy of risk perception subsequent to newborn genetic screening has been tested.

A second purpose of the study was to examine the role of dispositional optimism altruistic motives, information-seeking style, and satisfaction with information obtained as potential predictors of risk perception, anxiety, or stated intentions to participate in a longitudinal natural health study To ensure that mothers feel satisfied with the information they receive, and are well informed about their decision to continue or decline participation in a longitudinal natural health study of their at-risk child, it is important to determine what type of risk notification strategies work best for specific individuals

Based on the available published literature, the following *cognitive outcomes* are expected

(1) Accuracy of risk perception will change over time, with a decrease in those who are accurate and an increase in those who underestimate the child s risk

(2) Mothers from ethnic minority groups and mothers with lower levels of education will have less accurate risk perceptions

(3) Family history of diabetes of any type will be associated with increased accuracy of risk perception, both initially and over time

(4) Individualized letters reiterating what a mother was told over the phone regarding the child's risk will facilitate more accurate recall of the child s risk status compared to a no letter condition

(5) Mothers scoring high in information seeking are expected to have more accurate risk perceptions, both initially and across time

The following *affective outcomes* were hypothesized:

(1)Anxiety will be initially clinically elevated

(2) Mothers from ethnic minority groups and mothers with lower levels of education will report greater anxiety after being notified of the child's increased risk

(3) Higher actual infant risk will be related to higher levels of maternal anxiety

(4) Mothers scoring high in optimism will report lower levels of maternal anxiety than mothers lower in dispositional optimism

(5) Mothers scoring higher in information seeking will report higher levels of anxiety than mothers scoring lower in information seeking

(6) Mothers who report satisfaction with the amount of information they received will report less anxiety than those who are not satisfied

(7) The amount of information preferred and the amount of information obtained will interact, such that high levels of information seeking with low levels of satisfaction are expected to result in high levels of anxiety

(8) Mothers whose children have a family history of diabetes are expected to report more anxiety than mothers whose children have no family history of diabetes

(9) Over time, maternal anxiety levels will significantly decline to normal levels.

Finally, the following hypotheses were made regarding maternal *behavioral intentions* to continue participating in PANDA

(1) Caucasians will report greater intentions to continue in PANDA than African Americans

(2) Mothers of higher risk infants will report greater intentions to continue in PANDA than mothers of lower risk infants

(3) Mothers receiving an individualized follow-up letter post notification that provides a rationale for study participation (i.e., altruism or money) will be more likely to report intentions to continue in PANDA than those mothers who did not receive an individualized letter with an altruistic or monetary rationale for PANDA participation

(4) Anxiety is expected to be positively related to study participation, more anxious mothers will be more likely to report intentions to continue in PANDA.

(5) Mothers high in dispositional optimism will be less likely to report intentions to continue in PANDA

(6) Mothers with greater information-seeking behaviors will be more likely to report intentions to continue in PANDA.

(7) Over time, mothers with children who have a family history of diabetes will be more likely to reported intentions to continue in PANDA than mothers of children with no family history of diabetes

(8) Over time, the number of mothers willing to continue in PANDA will decline

CHAPTER 4
METHODS

Participants

Mothers of infants found to be at increased risk for the development of Type 1 diabetes through testing in PANDA were eligible for participation in this interview study. From January 2002 through May 2003, 273 mothers were notified of their child's risk status. Of the 273 mothers notified, 271 were eligible for participation in the interviews as two children were over three years of age at the time of notification. Of the 271 mothers, we were unable to reach 47 due to a disconnected phone or a failure to answer the phone after five attempts. Of the 210 mothers contacted, 190 agreed to participate in Follow Up Interview 1 (90.5%), which is consistent from previous findings with a similar population (Carmichael et al., 2003, Johnson, 2001). Mothers chose to participate in the interview even if they were deciding about (16.6%), or chose not to continue in (13%) the PANDA study. One hundred forty-two mothers were able to complete a second follow-up interview, which is 74.7% of those who completed the Follow-Up Interview 1. Thirty-seven of the original 271 mothers were unable to be contacted for the Follow-Up Interview 2, largely due to disconnected phone numbers or a failure to answer the phone after five attempts to reach the mother. Only 5.6% (11 mothers) verbally declined to participate in the Follow-Up Interview 2.

Procedures

Mothers of newborns were approached in the hospital by members of the PANDA staff, and permission was obtained to screen the child's blood to determine whether the child had HLA DQB1 alleles that placed them at an increased risk for the development of Type I diabetes. Mothers who agreed subsequently had their newborn's blood taken at the time of the PKU heel-stick testing, and it was screened to assess risk. Based on the DR alleles, DQ genotype, and first-degree relative status, the infant's risk for diabetes development was determined, and the child was placed into one of six categories. (Refer to Appendix A for a table of how infants are categorized based on genotype and family history of diabetes.) Mothers whose newborn was determined to be in either the low, very low, or protected risk categories were told they would not receive further contact from the PANDA study. Mothers of newborns found to be at increased risk (i.e., moderate, high, or very high risk) were contacted to request further participation in PANDA. Parents of at risk newborns were mailed a letter requesting they call the PANDA study coordinator to discuss the results and further participation in PANDA (refer to Appendix B for study design). If no response was received, mothers were contacted by PANDA staff to notify her of her newborn's risk. These children were on average, 301 days old when the mother was notified of the child's risk (range = 51-827 days, SD=146).

When the mother of the infant was notified, the study coordinator followed a scripted presentation of risk information, including the presentation of both categorical and numerical risk figures using both categorical terminology (i.e., moderate, high, extremely high) and numerical estimates (2%-25%) of risk, as suggested by the literature. For example, a portion of the script read, "We have found that your baby is what we would call at *moderate risk* for developing diabetes. Please keep in mind that this does not mean that your baby will definitely get diabetes Out of 100 babies with the same genetic markers as your baby there would be approximately 2 babies that would go on to develop diabetes" (see Appendix C for the full script) At the time of the Notification Interview, the following variables were recorded actual infant risk, date of notification, whether the mother or the study coordinator had initiated the notification phone call length of initial notification phone call in minutes and whether additional diabetes/PANDA information was requested by the mother

The mother was presented with the available options including further participation in PANDA If the mother requested additional information regarding PANDA, she was mailed a general information letter that broadly addressed Type 1 diabetes and the goals of the PANDA study This general information letter did not contain risk-specific information, a monetary reminder nor provide an altruistic rationale for participation

Parents had the option at the time of this Notification Interview of declining further participation, stating that they would like to continue with the study, or stating that they would like to delay their decision. Regardless of their decision about participation in PANDA, all mothers were asked for their permission to be contacted for a follow-up telephone interview

Immediately after the mothers were informed of the infant s at-risk status mothers were randomized to one of four groups stratified across actual infant risk group (moderate high, very high) One group received a tailored follow up risk information letter which simply restated the infant's risk in both categorical and numerical terms, modified for her child's specific risk A second group received an identical letter with tailored risk information plus an additional paragraph emphasizing an altruistic motive for participation, stating the child was "very special and may help to provide important information that may lead to the prevention of Type 1 diabetes" A third group received an identical tailored risk information letter plus a reminder about the $20 monetary compensation offered for their time and effort (refer to Appendices D and E for examples of letters sent) The final group received no letter at all

Allowing for at least one week for the letters to reach the families, mothers who agreed to the follow up interview were contacted by phone (mean length of time after notification of risk status=30 0 days SD 25 7 days) Follow up Interview 1 was conducted and the following variables assessed (see Appendix F for copy of Follow-Up Interview 1) risk perception socio-demographics including family history of diabetes; satisfaction with information received anxiety dispositional optimism, information-seeking behavior and behavioral intentions to continue in PANDA

Approximately 2.6 months (SD= 73 months) after Follow Up Interview 1, mothers were contacted for a Follow Up Interview 2 and the following variables re assessed (see Appendix G for Follow Up Interview 2) risk perception, anxiety, information seeking behavior satisfaction with information received and behavioral intentions to continue in PANDA

The University of Florida Health Science Center Institutional Review Board (IRB-01) reviewed the written proposal and approved the study as exempt from written informed consent as it was (1) minimal risk and (2) research on individual or group characteristics or behavior or research employing an interview

Measures

Maternal characteristics. Maternal characteristics were assessed during Follow-Up Interview I and are presented in Table 5.1. For the purposes of data analysis, the ethnic groups of Asian, Hispanic, and other were collapsed together due to small sample sizes. African American mothers constituted a larger group (N=26) and were able to be analyzed alone. If no effect was found for African-American status, the data were collapsed and examined as 1=Caucasian or 0= Non-Caucasian. Similarly, a conceptual categorical division was made for maternal education level (coded 1 if ≥ some college or 0≤ if high school high school or less), maternal marital status (coded 1=married or 0=single/divorced/separated). Maternal age was also assessed and used as a predictor variable.

Child characteristics. Based on the DR alleles, DQ genotype, and first-degree relative status, an infant risk status was recorded at the time of the Notification Interview. Three risk groups were eligible for the study: moderate, high, and extremely high risk. However, only one baby was in the extremely high risk group. Consequently, all babies' risk was coded 1=high or extremely high risk and 0=moderate risk. The child's family history of diabetes was assessed (coded 1= any family history of diabetes or 0=no family history of diabetes). For the purpose of data analysis, family history of diabetes was examined as having any history of diabetes, due to having only nine subjects with a first degree relative with diabetes. Child gender (coded 1=male or 0=female), and date of birth were recorded. Also assessed was whether or not this is the first child (coded 1=yes, first child or 0=no, not first child).

Intervention group. Mothers were randomized to one of four groups for the receipt of the intervention letter: tailored risk information letter alone, tailored risk information letter plus an altruistic rationale for study participation, tailored risk information letter plus a description of the monetary incentive the mother would receive for participation, or no tailored risk information letter of any type. Receipt of the letter was assumed. Mothers were not asked if they had received the letter since interviewers were blind to random assignment and such a question might raise anxiety or concerns for mothers who were not sent a letter. For the purposes of data analysis, data were examined as 1=tailored risk information intervention letter sent, any type or 0=no tailored risk information letter condition. When predicting to maternal intentions to continue with the PANDA study, the data were first examined for effect for type of tailored risk information letter, coded as 1= tailored risk information letter only vs. 0=no tailored risk information letter only; 1= tailored risk information + altruistic letter vs. no tailored risk information + altruistic letter, and 1=tailored risk information + monetary letter vs. 0=no tailored risk information + monetary letter. Since no significant effects for type of tailored risk information intervention letter were found, the data were collapsed together and examined as one variable coded 1=tailored risk information intervention letter sent or 0=no type of tailored risk information intervention letter sent.

Notification and interview dates. The date of the Initial Notification Interview was recorded. The dates of the two subsequent Follow-up Interviews were recorded to allow calculation of time elapsed since notification.

Satisfaction with information received. At both Follow-Up Interview 1 and Follow-Up Interview 2, mothers were asked to rate the amount of information they received about the study according to one of the three statements: "I received too little information about the PANDA study." "I received just the right amount of information about the PANDA study" or "I received too much information about the PANDA study." For the purposes of data analysis, satisfaction was coded as 1=satisfied with information received or 0=not satisfied with information received. Table 5-2 shows the percent of mothers who endorsed each statement at each interview.

Dispositional optimism. The Life Orientation Test (LOT; Scheier and Carver, 1985) is an eight-item scale designed to assess individual differences in generalized outcome expectancies, or dispositional optimism vs. pessimism. Of the eight items, four are scored in a positive direction and four are scored in a negative direction. During the course of Follow-Up Interview 1, mothers were asked to indicate the extent to which they agreed with each of the items, using the following response format: 4=strongly agreed, 3=agreed, 2=neutral, 1=disagreed and 0=strongly disagreed. Sample items included: "In uncertain times, I usually expect the best," and "I'm a believer in the idea that "every cloud has a silver lining." Items were reverse scored if appropriate and totaled to obtain a composite score. Scheier and Carver (1985) reported the scale has adequate internal consistency (.76); similar findings have been reported in other studies using the LOT (Carver et al., 1993). In the present sample, α= .82 (N=190), range= 14-40, mean 30.9, SD=5.09.

Information-seeking. The following objective information-seeking variables were recorded and coded from the Risk Notification Interview conducted by the PANDA study coordinator: how initial contact with the mother was initiated (1=mother called either prior to or after first letter stating results were available or 0=coordinator reached mother by phone after no response to letter stating results were available), length of the initial notification phone interview in minutes, and whether or not the mother requested to be sent additional general information about diabetes or the PANDA study (1=yes, general PANDA information requested or 0=no general PANDA information requested). In Follow-Up Interviews 1 and 2, a self-report measure of information-seeking was calculated using five yes/no items with one point assigned to each yes response. These items included: did you speak with a family member about your child's risk to gain information about your family history (1=yes or 0=no), talk to your pediatrician or other medical professional about your child's risk (1=yes or 0=no), call study personnel to discuss your child's risk (1=yes or 0=no), read a book on diabetes/diabetes risk (1=yes or 0=no), or search the Internet for information on your child's risk (1=yes or 0=no)? At Follow-Up Interview 1, the scale had rather poor internal consistency, coefficient alpha = .45 (N=190, M=1.35, SD=1.11) which increased to .60 (N=140, M=1.70, SD=1.36) at Follow Up Interview 2.

Accuracy of perceived risk for Type 1 diabetes. Mothers were asked at Follow-Up Interviews 1 and 2, "do you recall, what you were told regarding your child's risk for developing diabetes?" The mothers' spontaneous response was recorded. If no response was given, or only one category or number given, the interviewer prompted, "do you recall any categories/numbers?" The response was recorded, and if the mother could not recollect or was inaccurate in her recall, she was provided with an opportunity for recognition. Mothers were read a list of risks (both categorical and numerical) and asked whether or not any of those was the child's risk group. "I don't know" was recorded if the mother was unable to recall or recognize her child's risk category or number.

Accuracy of perceived risk was classified as "accurate", "overestimates", "underestimates", or "don't know". Perceived risk was considered accurate if the participant was able to either spontaneously recall or recognize the infant's correct risk status utilizing either a categorical or numerical estimate. "Overestimators" were classified as those mothers who recalled and/or recognized (after all opportunities are given) a risk figure (either categorical or numerical) that is higher than the actual risk figure told to the mother. "Underestimates" were identified as those mothers who recalled and/or recognized a risk figure lower than the infant's actual risk. Percent classified in each category at both Follow-Up Interviews 1 and 2 are reported in Table 5-4. For data analysis, accuracy was dichotomized into 1=accurate or 0=not accurate (included overestimators, underestimators, and those who did not know the risk). For a separate analyses of underestimators, data was coded as 1=underestimated risk or 0=did not underestimate risk or did not know.

Anxiety. The STAI-SF, a six-item Short Form of the state portion of the State Trait Anxiety Inventory (STAI, Spielberger, 1980) was previously developed and modified for use in this particular population (Johnson et al., in press). It assesses the participant's degree of anxiety about the child's risk for developing Type 1 diabetes. The six-item short form correlates highly with the full scale in this population (r=.95), and the regression equation used to predict to full scale anxiety scores will be described further below. The range for the full scale STAI is 20-80, with normative healthy female scores reported as 36.2 (SD=11) (Speilberger, 1980), placing clinically elevated cutoffs at 58, or 2 standard deviations above the mean. Normative data are provided in the STAI Manual, and other comparisons groups are available comparison full scale scores, including (1) mothers learning of their child's increased risk status as a result of ICA testing (Tercyak & Johnson, 1995) (2) pregnant women undergoing amniocentesis (Tercyak, Johnson, Roberts, & Cruz, 2001) and (3) a sample of mothers learning of their newborns genetic risk for developing Type I diabetes (Johnson et al., in press). In each case, the scale has been found to have high internal consistency.

Total scores for the six-item short form (SF) scale range from 4-24, with higher scores indicating greater anxiety. The regression equation predicting to the full scale was anxiety= (2.62 * STAI-SF) + 6.33. For this sample, the coefficient alpha for the STAI-SF at Follow-Up Interview 1 was α= .87 (N=190, M=36.04, SD=10.56) and α= .90 at Follow-Up Interview 2 (N=142, M=31.60, SD=10.45). The STAI-SF was used as a predictor of both accuracy of perceived risk and behavioral intentions to continue in the study.

Intentions to participate in PANDA. Intentions toward continued study participation were assessed at all three interviews (Notification Interview, Follow-Up Interview 1, Follow Up Interview 2) with a single item, "At the present time, which of the following statements best describes your feelings regarding your child's participation in this study: I am not considering continued participation in the PANDA study at this time, I am deciding whether or not my child will continue to participate in PANDA, I am definitely going to continue my child's participation in the PANDA study." Percent of mothers who endorsed each item may be seen in Table 4-4. For the purposes of data analysis, this outcome variable was dichotomized into 1=definitely will continue in PANDA and 0=undecided/definitely will not continue in PANDA.

CHAPTER 5
RESULTS

Demographic and Family Characteristics

Tables 5 1 and 5-2 describe the characteristics of the 190 mothers and infants participating in Follow-Up Interview 1 The mothers who participated in the interview study tended to be Caucasian (83 1%), married (70 7%) and had at least some college education (67 7%) Most of the infants were classified as in the moderate risk category (70 5%) A family history of diabetes of any type was reported in 130 (68.4%) of the families Further description of the maternal responses to variables assessed at the Risk Notification Interview, Follow Up Interview 1, and Follow Up Interview 2 may be viewed in Table 5-2

Comparisons of those who completed none vs one vs two interviews

A binary logistic regression analysis was used to examine the variables known at the Risk Notification Interview that may be related to interview completion. These characteristics included infant risk, type of tailored risk information letter group randomization, infant age at notification, length of notification call whether the mother or PANDA staff initiated the Risk Notification Interview, and number of days elapsed since notification. No variables were found to be predictive of mothers who participated in Follow-Up Interview 1 (N = 190) compared to those mothers who did not participate in Follow-Up Interview 1 (N= 67) Of those mothers who participated in Follow Up Interview 1 74 7% participated in Follow Up Interview 2 this is consistent with

Table 5.1. Demographic and Family Characteristics of Study Sample (N=190)

Maternal Characteristics	*N(%)*
Maternal Age at time of notification (years) M±SD	27.6 (6.2)
Maternal Ethnicity	
Caucasian	154 (81.1)
African American	26 (13.7)
Hispanic American	7 (3.6)
Asian American/Other	3 (1.6)
Marital Status	
Married	133 (70.0)
Single (father involved)	35 (18.4)
Single (father not involved)	13 (6.8)
Divorced/Separated	7 (3.7)
Did not report	2 (1.1)
Maternal Education	
Some High School	25 (13.2)
High School Degree	42 (22.2)
Some College	61 (32.3)
College Degree	41 (21.6)
Some Graduate School	2 (1.1)
Post-Graduate Degree	18 (9.5)
Did not report	1 (0.5)
Income	
0 – 20K	47 (25.0)
20 – 40K	60 (31.9)
40 – 60K	29 (15.4)
>60 K	42 (22.3)
Did not know/Did not report	12 (6.4)
Child Characteristics	
Child Age (days) at time of notification	301.14 (146.837)
Child Gender 3(% Female)	99 (46.8)
Risk Status	
Moderate Risk	134 (70.5)
High Risk	56 (29.5)
Only Child	
Yes	67 (62.1)
No	118 (35.3)
Did not report	5 (2.6)
Family History of Diabetes	
First degree relative	9 (4.7)
≥ Second degree relative	121 (63.6)
None	60 (31.6)

Table 5-2. Maternal Cognitive & Affective Variables

Variables	*Notification* *N (%)*	*Follow-Up 1* *N (%)*	*Follow Up 2* *N (%)*
Mom requested additional PANDA/Diabetes information?			
Yes	124 (65.3)	--	--
No	66 (34.7)	--	--
Who Initiated Notification Call			
Mother	26 (12.7)	--	--
Coordinator	147 (77.4)	--	--
Not Recorded	17 (8.9)	--	--
Length of Notification Phone Call M(SD)	4.46 (3.18)	--	-
Information-Seeking by Mother (%Yes)			
Spoke to family member	--	65 (35.5)	36 (25.9)
Spoke to medical personnel	--	42 (23.0)	29 (20.9)
Spoke to PANDA staff	--	23 (12.6)	27 (19.4)
Searched for information on Internet	--	5 (2.7)	12 (8.6)
Read a book or literature for info	--	2 (1.1)	3 (2.2)
Did none of the above	--	46 (25.1)	32 (23.0)
Composite Score M(SD)	--	1.30 (1.11)	1.75 (1.13)
How much information did you receive?			
Too much information	--	2 (1.1)	1 (.7)
Just the right amount of information	--	115 (61.2)	100 (71.4)
Not enough information	--	71 (37.8)	39 (27.9)
How do you feel about PANDA participation?			
Glad	--	176 (92.5)	127 (92.7)
Unsure	--	13 (6.8)	7 (5.1)
Wish we had not participated	--	1 (0.5)	3 (2.2)
Plans for PANDA?			
Yes we will continue in PANDA	116 (61.7)	131 (69.7)	90 (63.8)
Unsure about continuing	55 (29.3)	32 (17.0)	27 (19.1)
No, not continuing with PANDA	17 (9.0)	25 (13.3)	24 (17.0)
Dispositional Optimism M(SD)	--	30.09 (5.09)	--
State Anxiety M(SD)	--	36.04 (10.56)	31.60 (10.45)

previous findings for maternal participation with the Follow-Up Interview 2 (Carmichael et al., 2003, Johnson 2001). Comparisons between the mothers who participated in both Follow-Up Interviews (n = 142) and those mothers who participated in only the first

Follow Up Interview (n – 48) revealed differences in infant gender and dispositional optimism Mothers who completed both Follow Up interviews were more likely to have a male child (coded 1=male 0=female) (B= 709, p= 04) and were marginally more likely to report greater dispositional optimism than those interviewed only once (B= 058, p= 07) See Table 5-3 for group differences

Table 5-3. Differences between Follow Up Interview Completers and Drop-Outs

Variables	*Completed only 1 Follow Up Interview (N 48)*	*Completed 2 Follow-Up Interviews (N–142)*
Child Gender		
% Male	66 7%	49 6%
Dispositional Optimism	29 6 (5 4)	31 1(5 0)

Note Coded as 1–Completed multiple interviews or 0=completed one interview

Data Analysis Approach

Missing data values were replaced with sample means for each item missing for a small number of subjects who were missing one or two items on the STAI, LOT, or Information Seeking data at Follow-Up Interview 1 No linear data were missing at Follow-Up Interview 2 and missing categorical data were not replaced at any time point.

Hierarchical binary logistic regression models were used to examine predictors of maternal accuracy for both follow up interviews For entry into the regression equations, variables were grouped in conceptually meaningful blocks and entered into the models in an a priori order, in this case, more general variables were entered earlier (e g , maternal demographic characteristics) and variables expected to be more closely related to the dependent variable of interest, such as maternal information-seeking were entered later These blocks included maternal characteristics (maternal age, marital status maternal education, maternal ethnicity), child characteristics (child age, only child status number of children in family, family history of diabetes, infant risk status), intervention variables (tailored risk information letter vs. no letter, length of time since notification), maternal affective variables (maternal anxiety, maternal dispositional optimism, maternal satisfaction), objective maternal information-seeking variables (length of notification phone call, who initiated risk notification interview, whether mothers requested generalized information letter) and subjective maternal information-seeking (maternal information-seeking composite). For those regression equations predicting to the maintenance of maternal accuracy over time (e.g., from Follow Up Interview 1 and Follow-Up Interview 2), Follow Up Interview 1 maternal accuracy was first entered into the regression equation, followed by the hierarchical entry of variables in the blocks as described. Only those variables that met the $p < .10$ criterion for retention in each block were kept for subsequent analysis.

Change in maternal accuracy over time was examined using a McNemar chi-square test for dependent data. Only those mothers who completed both interviews were included in this analysis.

Accuracy of Maternal Risk Perception

The first major aim of this study was to determine the relative roles of differing risk notification strategies, dispositional optimism, ethnicity, and educational level, information-seeking and satisfaction with information obtained as potential predictors of the accuracy of maternal perception of the newborn's risk for diabetes.

Accuracy of Recall

During Follow Up Interview 1 and Follow-Up Interview 2, mothers were asked to recall the at-risk status of their newborn. If they were unable to recall the at-risk status spontaneously, they were given prompts regarding the verbal label and/or the numerical label for the newborn's risk category. Mothers were identified as *accurate* if they were able to accurately report the at-risk category and/or numerical value of the newborn's risk category. This could have occurred on either the recall or recognition task or both. Mothers who were not accurate were identified as *overestimators* if they reported a higher level of risk than actual risk (e.g., reported High risk when newborn was actually Moderate risk) or they were identified as *underestimators* if they reported a lower level of risk than actual risk (e.g., reported Moderate risk when newborn was actually High risk). Finally, mothers who did not remember or did not know the risk category were identified as mothers who *did not know*.

Change in Accuracy Over Time

Mothers who completed Follow-Up Interview 1 were largely accurate in their recall. Of 190 mothers, 148 (77.9%) were accurate in reporting their newborn's risk category, number, or both (see Table 5-4). At Follow-Up Interview 2 the percentage of mothers who were accurate decreased and the percentage who underestimated the child's risk increased. Using a McNemar chi square analysis for dependent data, the percent change from 77.9% to 69.0% of mothers who remained accurate was significantly different at the p=.003 level (χ^2=4.69, p=.003).

We also examined the change for those mothers who underestimated their child's risk over time and found a significant effect (χ^2=4.69, p=.035), indicating an increase in the number of mothers who underestimated their child's risk over time. The change in overestimation or in mothers who did not know over time was not statistically evaluated because of the very small number of mothers who fell in these categories.

Table 5-4. Accuracy of Recall of Risk

	Follow-Up Interview 1 (N = 190)		Follow-Up Interview 2 (N = 142)	
Recall	Number	%	Number	%
Accurate	148	77.9	98	69.0
Underestimate	23	12.1	27	19.0
Overestimate	2	1.1	3	1.6
Don't know or remember	17	8.9	14	9.9

Predictors of Accuracy of Risk Perception at Follow-Up Interview 1

Binary categorical logistic regression was used to examine predictors of maternal accuracy for both interviews. As outlined, variables were entered in blocks consisting of maternal characteristics in block 1, child characteristics in block 2, intervention variables in block 3, maternal affective variables in block 4, objective maternal information-seeking variables in block 5 and subjective maternal information-seeking report in block 6. At each step, variables were retained if $p < .10$. Table 5-5 describes the results of this analysis.

Results indicated that maternal educational level, the tailored risk information letter intervention, length of notification phone call, and maternal report of information seeking was predictive of maternal accuracy of newborn risk perception at the Follow-Up Interview 1. Mothers who were more accurate about their child's risk during Follow-Up Interview 1 had some college education or greater, were randomized to receive a post-notification tailored risk information letter re-stating their child's specific risk status, had a longer notification phone call, and reported using more attempts to gain information after notification regarding their child's risk for developing diabetes, either through family, medical professionals, or media. The model was significant at $\chi^2=17.73$, $p=.001$, Cox and Snell $R^2=.106$.

Table 5-5 Logistic Regression predicting to Mothers who are Accurate vs. Not Accurate Follow-Up Interview 1 (N=190)

Variable	*B*	*SE B*	*p*
Block 1-Maternal characteristics			
Maternal education 1 ≥ college, 0≤ high school	716	359	046
Block 2-Child Characteristics			
No additional predictors	716	359	046
Block 3-Intervention Variables			
Maternal education	780	365	033
Tailored risk information letter, any type 1 =tailored letter sent, 0=tailored letter not sent	670	385	082
Block 4-Maternal Affective Variables			
Maternal education	958	386	013
Tailored risk information letter	739	391	059
Maternal anxiety at Follow-Up Interview 1	031	019	098
Block 5-Objective Maternal Information-Seeking Variables			
Maternal education	833	.393	034
Tailored risk information letter	875	407	032
Maternal anxiety at Follow Up Interview 1	029	019	126
Length of notification phone call in minutes	269	117	.022
Block 6-Subjective Maternal Information-Seeking			
Maternal education	661	376	079
Tailored risk information letter	709	405	080
Length of notification phone call in minutes	.266	114	020
Information Seeking at Follow-Up Interview 1	328	191	086
Final model			
Maternal education	661	376	079
Tailored risk information letter	709	405	080
Length of notification phone call in minutes	266	114	020
Information Seeking at Follow-Up Interview 1	328	191	086

Note Accuracy coded as 1=Accurate risk estimate and 0=Non-Accurate risk estimate

Predictors of Accuracy Over Time

In order to determine those factors contributing to the maintenance of accurate risk estimates across time a separate binary logistic regression model was used with Follow-Up Interview 2 accuracy as the criterion Accuracy at Follow-Up Interview 1 was entered first in the regression model, with additional predictor variables entered in blocks as previously described

The final model included Follow-Up Interview 1 accuracy, whether or not the mother had been assigned to receive one of the three types of risk information letters post notification (with either information only, altruistic plea, or monetary reminder), Follow-Up Interview 2 maternal anxiety, and whether or not the mother specifically requested general PANDA information at the time of notification (letter sent did not include specific risk information). Refer to Table 5-6. Mothers who were initially accurate in their report of the newborn's risk status at the Follow-Up Interview 1 were more likely to be accurate during the Follow-Up Interview 2. More accurate mothers at the Follow-Up Interview 2 also had received a tailored risk information letter reiterating their own child's risk, requested additional general information regarding PANDA, and reported higher current anxiety than mothers who were not accurate in their risk recall at Follow-Up Interview 2. The model was significant at $\chi^2=30.40$, $p=.001$, Cox and Snell $R^2=.194$.

Underestimation of Newborn Risk

Next, a binary logistic regression analysis was conducted for the group of mothers who reported an underestimate of their child's risk status at Follow-Up Interview 1. For this analysis, mothers who underestimated their child's risk were categorized as "underestimators," placing the mothers who were accurate, overestimators, or who did not recall/know into the category of "non underestimators." A similar pattern of variable entry was used in this regression analysis as in prior regression analyses.

Number of days since risk notification, maternal anxiety, length of notification phone call, and whether or not the mother requested additional generalized written PANDA/diabetes informational materials were associated with underestimation of risk at

Table 5-6 Logistic Regression for Follow-Up Interview 2 Accuracy (N=142)

Variable	*B*	*SE B*	*p*
Block 1 Prior interview characteristics			
Maternal Accuracy at Follow-Up Interview 1 1 Accurate 0=Not Accurate	1 782	0 442	000
Block 2 Maternal characteristics			
No additional predictors	1 782	0 442	000
Block 2-Child Characteristics			
Maternal accuracy at Follow-Up Interview 1	1 870	459	000
Child risk status 1=High 0=Moderate Risk	1 036	484	032
Block 3-Intervention Variables			
Maternal accuracy at Follow-Up Interview 1	1 835	468	000
Child risk status 1=High 0=Moderate Risk	1 061	487	029
Tailored risk information letter, any type 1 =sent, 0=not sent	827	448	065
Block 4-Maternal Affective Variables			
Maternal accuracy at Follow-Up Interview 1	1 848	475	000
Child risk status 1=High 0=Moderate Risk	870	504	084
Tailored risk information letter, any type 1 =sent, 0=not sent	873	457	056
Maternal anxiety at Follow-Up Interview 2	031	022	153
Block 5-Maternal Objective Information-Seeking Variables			
Maternal accuracy at Follow Up Interview 1	2 100	510	000
Child risk status	790	513	124
Tailored risk information letter	1 958	720	007
Maternal anxiety at Follow-Up Interview 2	039	023	096
General PANDA information letter requested 1=yes 0=no	1 467	704	037
Block 6- Maternal Subjective Information-Seeking			
No additional predictors			
Final Model			
Maternal Accuracy at Follow-Up Interview 1	2 068	501	.000
Tailored risk information letter	1 981	710	005
Maternal anxiety at Follow-Up Interview 2	050	023	029
General PANDA information letter	1.543	698	027

Note. Accuracy coded as 1=Accurate risk estimate and 0=Non-Accurate risk estimate

Follow-Up Interview 1 Underestimators had a greater amount of time since risk notification, reported less anxiety and more satisfaction with the amount of information received and were more likely to request generalized PANDA information than non-underestimators The model was significant at $\chi^2=16.67$, $p=.002$, Cox and Snell $R^2=.087$

Final results are presented in Table 5 7

Table 5 7 Logistic Regression for predicting Underestimators vs. Non-Underestimators at the Follow-Up Interview 1 (N=190)

Variable	*B*	*SE B*	*p*
Blocks 1& 2 Maternal & Child Characteristics			
No predictors			
Block 3 Intervention Variables			
Days elapsed since notification of risk	0.012	0.007	.071
Block 4-Maternal Affective Variables			
Days since risk notification	.013	.007	.070
Satisfaction with information received 1=Satisfied 0=not satisfied	1.013	.535	.058
Maternal anxiety at Follow-Up Interview 1	-.053	.025	.034
Blocks 5 Objective Maternal Information-Seeking			
Days since risk notification	.016	.007	.036
Satisfaction with information received	1.088	.544	.046
Maternal anxiety at Follow-Up Interview 1	-.062	.026	.020
General PANDA information requested	1.066	.482	.027
Block 6-Subjective Maternal Information Seeking			
No additional predictors			
Final Model			
Days since risk notification	.016	.007	.036
Satisfaction with information received	1.088	.544	.046
Maternal anxiety at Follow Up Interview 1	-.062	.026	.020
General PANDA information requested	1.066	.482	.027

Note Outcome variable coded as 1=Underestimators and 0=all other risk estimates.

Predictors of Underestimation Over Time

A second binary logistic regression analysis was then conducted for Follow-Up Interview 2 underestimators. In the initial block of the analysis, Follow Up Interview 1 underestimator status was entered Additional predictor variables retained in the model included African American maternal ethnicity and maternal anxiety at the time of Follow-Up interview 2 Mothers who were more likely to underestimate their newborn's risk over time had underestimated risk initially, were African-American and reported lower anxiety levels than those mothers who did not underestimate their infant's risk The model was significant at χ^2=28 78 p=000, Cox and Snell R^2=185 Results are presented in Table 5 8

Table 5-8. Logistic Regression for Underestimation at Follow-Up Interview 2 (N=139)

Variable	*B*	*SE B*	*p*
Block 1-Initial interview variables			
Underestimators at Follow Up Interview 1 1=underestimated risk 0=did not underestimate	2 350	579	000
Block 2 Maternal characteristics			
Underestimators at Follow-Up Interview 1	2 629	606	000
Maternal African American Ethnicity 1=African American 0=other	1 527	581	009
Block 3-Child Characteristics			
Underestimators at Follow-Up Interview 1	2 946	666	000
Maternal African American Ethnicity	1 531	598	010
Child Gender 1=male, 0=female	904	915	078
Block 4 Intervention Variables			
No additional predictors			
Block 5 Maternal Affective Variables			
Underestimators at Follow-Up Interview 1	2 686	634	000
Maternal African American Ethnicity	2 146	680	002
Maternal Anxiety at Follow Up Interview 2	- 057	028	043
Blocks 5 6 & 7 Maternal Cognitive Information Seeking Behavior-Objective/Subjective			
No additional predictors			
Final Model			
Underestimators at Follow-Up Interview 1	2 686	634	000
Maternal African American Ethnicity	2 146	680	002
Maternal Anxiety at Follow-Up Interview 2	057	028	043

Note Outcome variable coded as 1=Underestimators and 0=all other risk estimates

Summary of Outcomes Related to Accuracy of Maternal Risk Perception

Four of the five hypotheses related to maternal accuracy of risk perception were supported Nearly 80% of women were accurate in their initial recall of risk, with more women underestimating infant risk over time Mothers were more likely to be accurate about their child's risk if they had higher levels of education, had a longer notification phone call, received a tailored risk information letter, and if they used more information-seeking strategies to find out about diabetes and diabetes risk Over time, mothers remained more accurate if they were accurate initially had received a tailored risk information letter reiterating the child's risk, had requested additional generalized informational materials regarding the PANDA study, and reported high anxiety

Mothers were more likely to underestimate their child's risk if they had a longer length of time since notification reported lower levels of anxiety but more satisfaction with information received, and if they had requested information materials regarding the PANDA study Over time, underestimators were more likely to have underestimated at Follow Up interview 1 identify with an African American ethnicity, and report lower levels of anxiety than those who did not underestimate infant risk

The following hypotheses supported or partially supported include 1) Accuracy of risk perception will change over time, with a decrease in those who are accurate and an increase in those who underestimate the child's risk, 2) Mothers from ethnic minority groups and those with lower levels of education will have less accurate risk perceptions. 3) Tailored risk information letters reiterating what a mother was told over the phone regarding the child's risk will facilitate more accurate recall of the child's risk status compared to a no letter condition and 4) Mothers scoring higher in information-seeking will have more accurate risk perceptions, both initially and across time The data did not support the hypothesis that a history of diabetes of any type was associated with increased accuracy of risk perception

Maternal Anxiety in Response to Risk Notification

Statistical Analyses

Anxiety was measured by the six-item STAI SF at Follow-Up Interview 1 and Follow-Up Interview 2. A regression equation previously calculated from the sample used to develop the STAI-SF was used to predict from the STAI-SF to the full scale STAI, for comparative purposes (Anxiety= 2.62SF + 6.33). Welch's T-tests were used to compare maternal STAI scores with normative data and STAI scores from comparison groups.

Hierarchical linear regression models were used to examine predictors of STAI scores for each of the three interviews. As before, more general, demographic variables were entered earlier in the model and variables expected to be more closely related to the mother's STAI scores (e.g. optimism) were entered later. When predicting STAI scores at Follow Up Interview 1, the blocks to be entered included maternal characteristics (maternal age, marital status, maternal education, maternal ethnicity), child characteristics (child age, only child status, number of children in family, family history of diabetes, infant risk status), intervention variables (tailored risk information letter vs no letter, length of time since notification), maternal affective variables (maternal dispositional optimism, maternal satisfaction), objective maternal information-seeking variables (length of notification phone call, who initiated risk notification interview, whether mothers requested a generalized information letter) and subjective maternal information-seeking (maternal information-seeking composite). For prediction of maternal anxiety over time (e.g., from Follow Up Interview 1 and Follow-Up Interview 2), Follow-Up Interview 1 maternal accuracy was first entered into the regression equation, followed by the hierarchical entry of variables in the blocks as described. Only those variables that met the $p < .10$ criterion for retention in each block were kept for subsequent analysis.

Change in maternal anxiety over time was examined using a repeated measures analysis of variance. Between subjects variables included those variables that proved to be significant predictors of anxiety scores in the hierarchical regression analysis at Follow-Up Interview 1. The within-subjects variable was time of interview (Follow-Up Interview 1 vs. Follow Up Interview 2). Only those mothers who completed both interviews were included.

Comparisons with Normative and ICA+ Samples

Welch's T tests (for unpaired data assuming differing standard deviations) were used to compare maternal STAI scores with normative data (see Table 5-9). Mothers of genetically at-risk newborns reported STAI scores significantly lower than that of mothers of ICA+ children (*Tw*=-7.35, p<.001), and pregnant women undergoing amniocentesis (*Tw*=-7.99, p<.001), and not significantly different than pregnant women, working women, or mothers from a prior sample of newborns at-risk for Type 1 diabetes.

Predictors of Maternal Anxiety at Follow-Up Interview 1

Hierarchical linear regression models were used to examine predictors of STAI scores for both interviews by means of the same step-wise progression as described previously. An interaction term (information-seeking and maternal satisfaction) was added in the final block of the prediction model. Results indicated that mothers reporting more anxiety at Follow-Up Interview 1 tended to be African American, had an infant at high risk, had lower levels of dispositional optimism, and reported a greater number of

Table 5-9 Maternal STAI scores at Follow Up Interview 1 compared to parents of ICA+ children, pregnant women, and working women M±(SD)

	Mothers in current PANDA N=190	*Mothers in prior PANDA study*^{A} *N=184*	*Mothers of ICA+ children*^{B} *N=33*	*Pregnant women having Amnio*^{C} *N=200*	*Pregnant women*^{D} *N=200*	*Working women*^{E} *N=210)*
Total Sample ^{B,C}	36.1±10.5	36.04±10.7	55.4±14.4	44.9±11	37.6±11	36.2±11
(N)						
Moderate Risk (134)^{A,B,C,D,E}	33.9±9.9					
High Risk (56)^{A,B,C,D,E}	41.0±10.3					
African American (26)^{B,D,E}	44.2±11.0					
Non African-American (164)^{B,C,D}	34.7±9.9					
High in Optimism (107)^{A,B,C,D,E}	33.5±9.4					
Low in Optimism (80)^{A,B,C,D,E}	39.3±11.1					
High in Information Seeking (72)^{A,B,C,D,E}	39.3±10.6					
Low in Information Seeking (111)^{A,B,C,D,E}	33.7±10.1					

Note Data are means ± SD (N)

^{A} from Johnson & Tercyak (1995)

^{B} from Tercyak, Johnson Roberts & Cruz (2001)

^{C} from Marteau & Bekker (1992)

^{D} from Spielberger (1983)

^{E} from Johnson et al (in press)

information-seeking behaviors than mothers who had lower levels of anxiety An examination of the hypothesized interaction between information-seeking and satisfaction with information received did not reveal any effects on anxiety Refer to Table 5-10 for the model summary

Table 5-10 Follow-Up Interview 1 STAI Regression Analysis (N=190)

Variable	*B*	*SE B*	*β*	*p*	*Adj. R^2*
Block 1 Maternal characteristics					
Maternal education 1 ≥ college 0≤ high school	-3.776	1.533	-.171	.015	.122
Maternal African American ethnicity 1=African American 0=other	8.909	2.131	.290	<.000	
Block 2 Child Characteristics					
Maternal education	-3.772	1.442	-.168	.011	.232
Maternal African American ethnicity	9.632	1.993	.314	<.000	
Family History of Diabetes 1 ≥ first degree or greater 0=none	2.968	1.482	.130	.047	
Child Risk 1=High Risk 0=Moderate	7.276	1.508	.313	<.000	
Block 3 Intervention Variables					
No significant variables					
Block 4 Maternal Affective Variables					
Maternal African American ethnicity	10.028	1.910	.327	.000	.274
Child Risk	7.331	1.448	.317	.000	
Family History of Diabetes	2.595	1.415	.115	.068	
Dispositional Optimism	-.544	.129	-.263	.000	
Block 5 Maternal Objective Information Seeking					
No significant predictors					
Block 6-Subjective Information-Seeking					
Maternal African American Ethnicity	9.450	1.924	.308	.000	.277
Child Risk	7.423	1.439	.321	.000	
Dispositional Optimism	-.527	.129	-.254	.000	
Information-Seeking Time 1	1.210	.595	.127	.043	
Final Model					
Maternal African American Ethnicity	9.450	1.924	.308	.000	.277
Child Risk	7.423	1.439	.321	.000	
Dispositional Optimism	-.527	.129	.254	.000	
Information Seeking Time 1	1.210	.595	.127	.043	

Predictors of Anxiety at Follow-Up Interview 2

In order to examine those factors related to anxiety at Follow-Up Interview 2, Follow-Up Interview 1 STAI scores were entered first into a linear regression analyses, followed by the hierarchical entry of variables in blocks as described previously, including maternal satisfaction, information seeking, and the interaction of the two in the final block. Results of the final model accounted for nearly 38% of the variance in anxiety across time The final model (see Table 5-11) indicated that mothers who were more anxious at the Follow-Up Interview 2 Interview reported higher initial reports of anxiety were younger had higher risk newborns, reported lower levels of dispositional optimism, and had engaged in more information-seeking behavior than mothers who reported declining levels of anxiety

Change in Anxiety Over Time

Change in maternal anxiety over time was examined using a repeated measures analysis of variance Between subjects variables included those variables that proved to be significant predictors of maternal accuracy at Follow-Up interview 1 in the hierarchical regression analysis maternal African American ethnicity (coded as "African American" or "Non-African American"), infant risk ("high risk" vs "moderate risk"), dispositional optimism and information-seeking (both dichotomized as "high" or "low") The within subjects variable was time of interview (Follow Up Interview 1 Follow Up Interview 2) Only those mothers who completed both interviews were included As expected, maternal anxiety declined over time Only the main effect for time was significant [F(1)=8 76, p= 004]

Summary of Anxiety Outcomes

To summarize results with respect to maternal anxiety, we found that some mothers of at-risk infants reported anxiety levels that were significantly different from that of a normative sample, including African American mothers and mothers of infants identified as being at High Risk and these anxiety levels declined significantly over time Predictors of maternal anxiety at Follow Up Interview 1 found that mothers who reported higher levels of anxiety were African American, had an infant at high risk, reported lower

Table 5-11 Follow-Up Interview 2 STAI Regression Analysis (N=142)

Variable	B	SE B	β	p	Adj. R^2
Block 1-					
Follow-Up Interview 1 Maternal Anxiety	545	070	550	000	.298
Block 2 Maternal Characteristics					
Follow Up Interview 1 Maternal Anxiety	500	070	505	000	335
Maternal age	- 295	093	- 202	004	
Block 3-Child Characteristics					
Follow Up Interview 1 Maternal Anxiety	454	071	459	000	.356
Maternal age	364	116	- 217	002	
Child Risk 1=High Risk 0=Moderate Risk	3 798	1 591	167	018	
Block 4 Intervention Variables					
No additional predictors					
Block 5 Maternal Affective Variables					
Follow-Up Interview 1 Maternal Anxiety	416	075	420	000	364
Maternal age	- 324	118	- 194	007	
Child Risk	3 924	1 584	173	014	
Dispositional Optimism	- 252	156	- 119	107	
Block 6- Maternal objective Information Seeking –no additional predictors					
Block 7-Subjective Maternal Information-Seeking/Final Model					
Follow-Up Interview 1 Maternal Anxiety	374	076	.381	000	378
Maternal age	-.345	116	-.208	004	
Child Risk	3 621	1.594	161	025	
Dispositional Optimism	264	153	-127	087	
Maternal Information Seeking at Follow-Up Interview 2	877	543	115	109	

levels of dispositional optimism and engaged in more information-seeking behaviors than mothers reporting lower levels of anxiety Those who remained anxious over time tended to report higher initial anxiety, were younger, had an infant at increased risk, reported lower levels of dispositional optimism, and reported engaging in more information seeking behaviors

The results of the analysis supported the following hypotheses 1) Anxiety will be initially clinically elevated in some populations, 2) Mothers from ethnic minority groups will report greater anxiety after being notified of the child's increased risk, 3) Higher actual infant risk will be related to higher levels of maternal anxiety; 4) Mothers scoring higher in optimism will report lower levels of maternal anxiety than mothers lower in dispositional optimism 5) Mothers scoring higher in information seeking will report higher levels of anxiety than mothers scoring lower in information seeking, and 6) Over time, maternal anxiety levels will significantly decline to normal levels.

The data did not support the following hypotheses 1) Mothers with lower levels of education will report greater anxiety after being notified of the child's increased risk, 2) Mothers who stated satisfaction with the amount of information they received will report less anxiety than those who were not satisfied with the amount of information they received, 3) The amount of information preferred and the amount of information obtained will interact, such that high levels of information seeking with low levels of satisfaction result in higher levels of anxiety, and 4) Mothers whose children have a family history of diabetes will report more anxiety than mothers whose children have no family history of diabetes.

Behavioral Outcomes-Intentions to Continue with PANDA

Statistical Analyses

Behavioral intentions to continue with the PANDA Part II study were assessed at all three interviews (Notification interview Follow-Up Interview 1 and Follow-Up Interview 2) and were characterized as continuing with participation in PANDA, deciding about participation, or not continuing in the PANDA study For the purposes of regression analysis, the data was re-coded dichotomously as "definitely continuing" as compared to "not continuing/deciding"

Hierarchical binary logistic regression models were used to examine predictors of maternal intentions to participate in PANDA at all three interviews (Risk Notification

Interview, Follow-Up Interview 1 and Follow-Up Interview 2). Similar to before, for entry into the regression equations, more general variables were entered in earlier blocks and variables expected to be more closely related to maternal intentions were entered later. These blocks included maternal characteristics (maternal age, marital status, maternal education, maternal ethnicity), child characteristics (child age, child gender, only child status, number of children in family, family history of diabetes, infant risk status), intervention variables (tailored risk information letter vs. no letter, length of time since notification), maternal affective variables (maternal anxiety, maternal dispositional optimism, maternal satisfaction), objective maternal information-seeking variables (length of notification phone call, who initiated risk notification interview, whether mothers requested generalized information letter), subjective maternal information-seeking (maternal information-seeking composite), and maternal accuracy of risk estimation (mothers accurately recalled risk, or mothers underestimated child's risk).

For the regression equations predicting to the maintenance of maternal intentions to participate over time (e.g., from Risk Notification Interview to Follow-Up Interview I and Follow-Up Interview 2), the prior report or reports of maternal intentions was initially entered into the regression equation, followed by the hierarchical entry of variables previously listed in blocks. Once again, only those variables that met the $p < .10$ criterion for retention in each block were kept for subsequent analysis.

Change in maternal intentions to participate over time was examined using a McNemar chi-square test for dependent data. Only those mothers who completed both interviews were included in this analysis.

Maternal Intentions for PANDA Participation, Change Over Time

The majority of the 210 mothers (61.9%) indicated they intended to continue with the PANDA study at the time of notification, where as 29.3% were undecided and 9.0% indicated a desire for no further participation. Results of a McNemar chi square analysis for dependent data indicated that by the time of the Follow-Up Interview 1, significantly more (69.7%) mothers had agreed to participate in PANDA (χ^2=4.50, p=.034) which dropped to 63.8% at Follow-Up Interview 2. The number of mothers reporting indecision also significantly changed from 29.3% at the time of notification to only 17.0% at Follow-Up Interview 1 (χ^2=9.132, p=.003) and 19% at Follow-Up Interview 2. Upon initial notification, 9.0% of mothers indicated no desire to participate further in PANDA, which increased to 13.3% at Follow-Up Interview 1 and rose again at Follow-Up Interview 2 to 17.0%.

Predictors of Maternal Intentions for PANDA Participation

To determine the factors related to intention on continuing with the PANDA study, three binary logistic regression analyses were conducted in similar fashion to prior logistic regression analyses to predict stated intent to continue with the PANDA study at all three assessments. For the initial analysis, only known study variables could be utilized as predictors, including infant risk, infant gender, age of child at notification, whether the mother or study coordinator initiated the notification phone call, length of notification phone call, tailored risk information letter intervention group, and whether the mother requested additional PANDA information. None of these variables were found to be significantly related to whether or not a mother indicated a desire to continue to participate in PANDA at the notification interview.

Upon examining variables related to maternal intentions to participate in PANDA at the time of Follow-Up Interview 1, analysis indicated that mothers were more likely to state intentions to continue in the PANDA study at the time of the Follow-Up Interview 1 if they had previously stated such intentions to participate, if they reported having any family history of diabetes, and if they were less likely to underestimate of their infant's risk at the time of the Follow-Up Interview 1. The model was significant at χ^2=40.70, p=.000. Cox and Snell R^2=.196. (Refer to Table 5-12)

Analysis of the variables related to maternal intentions to participate in PANDA at the time of Follow-Up Interview 2 found that mothers were more likely to state intentions to continue in the PANDA study at the time of the Follow-Up Interview 2 if they had previously stated such intentions to participate at Follow-Up Interview 1 (but not at the Notification Interview), they identified with a non-Caucasian ethnicity, and if they had received a tailored risk information letter. Classification rate of the final model improved from 64.0% to 87.1%. The model was significant at χ^2=81.91, p=.002, Cox and Snell R^2=.445.

Table 5-12. Logistic Regression for Intent to Continue with PANDA study at time of Follow-Up Interview 1 (N=187)

Variable	*B*	*SE B*	*p*
Intent to Continue at Notification 1=intend to continue in PANDA 0=undecided/not continuing in PANDA	2.080	0.376	.000
Positive family history of diabetes 1=any family history 0=no family history	.791	0.384	.039
Maternal Underestimate of Risk 1=Underestimated 0=Did not underestimate	-1.098	.529	.038

Note. Intent to continue coded as 1. All other intentions regarding participation coded 0.

Table 5-13 Logistic Regression for Intent to Continue with PANDA study at time of Follow-Up Interview 2 (N=139)

Variable	B	SE B	p
Maternal Intentions to Participate at Follow-Up Interview 1	4.608	.805	.000
Maternal Ethnic Status is Caucasian 1=Caucasian 0=all other	-3.118	.988	.002
Tailored risk information letter 1=yes letter sent 0=no letter sent	1.066	.575	.064

Note. Intent to continue coded 1. All other intentions regarding participation coded 0.

Summary of outcomes related to maternal intentions for PANDA participation

There was no variable that predicted whether or not a mother would say she intended to continue from variables known at the notification interview. At the time of Follow-Up Interview 1, mothers who were more likely to state intentions to participate had previously stated intentions to participate at the notification interview, reported a positive family history of any type of diabetes, and were less likely to underestimate their infant risk compared to those mothers who did not state intentions to continue. Over time mothers who remained interested in participating in PANDA had expressed prior intent in PANDA participation, were more likely to be an ethnic minority mother, and had been randomized to receive a tailored risk information letter post-notification interview.

Results of data analysis found support for the following hypotheses: 1) Mothers receiving a tailored risk information letter post-notification will be more likely to report intentions to continue in PANDA than those mothers who did not receive an tailored risk information letter; 2) Mothers with children who have a family history of diabetes will be more likely to report intentions to continue in PANDA than mothers of children with no family history of diabetes; 3) Mothers who underestimated their child's risk will be less likely to report intentions to continue in PANDA, and 4) Over time, the number of mothers willing to continue in PANDA will decline

Hypotheses not supported included 1) Caucasians will report greater intentions to continue in PANDA than African-Americans, 2) Mothers of higher risk infants will report greater intentions to continue in PANDA than mothers of lower risk infants; 3) More anxious mothers will report intentions to continue in PANDA, 4) Mothers higher in dispositional optimism will be less likely to report intentions to continue in PANDA, and 5) Mothers with greater information-seeking behaviors will be more likely to report intentions to continue in PANDA.

CHAPTER 6
DISCUSSION

Review of Findings and Conclusions

Maternal Understanding of Newborn Risk

As in prior studies of PANDA mothers (Carmichael et al , 2003 Johnson, 2001) mothers' understanding of newborn risk was investigated in this study The first major aim was to determine the relative roles of differing risk notification strategies, dispositional optimism ethnicity, educational level, anxiety, information-seeking styles and satisfaction with information obtained as potential predictors of the accuracy of maternal perception of the newborn's risk for diabetes.

The following hypotheses were supported or partially supported 1) Accuracy of risk perception will change over time, with a decrease in those who are accurate and an increase in those who underestimate the child's risk, 2) Mothers from ethnic minority groups and those with lower levels of education will have less accurate risk perceptions, 3) Tailored risk information letters reiterating what a mother was told over the phone regarding the child's risk facilitated more accurate recall of the child's risk status compared to a no letter condition, and 4) Mothers scoring higher in information seeking had more accurate risk perceptions, both initially and across time The data did not support the hypothesis that a history of diabetes of any type was associated with increased accuracy of risk perception.

Supported hypothesis 1. Accuracy of risk perception will change over time, with a decrease in those who are accurate and an increase in those who underestimate the child's risk. It was confirmed that a majority of mothers were accurate in their understanding of the newborn's risk status initially, but accuracy declined over time. Further, underestimation of risk increased over time, accounting for the changes across time in overall accuracy. The rate of overestimation was small during both interviews. Previous research in our laboratory has also found an increase in those who underestimate the child's risk over time (Carmichael et al., 2003, Johnson, 2001). One of the conclusions regarding initial underestimation is that as time grows from the time of notification, there is a greater tendency to underestimate the newborn's risk. This is consistent with the many studies which have demonstrated that recipients of genetic information tend to become less accurate over time (e.g., Axworthy et al., 1996, Kessler, 1989, Ormond, Pergament, & Fine, 1996). Gordern, Walpole, Zubrick, & Bower (2003) attribute imprecise recall to memory loss, simplification of the result meaning, and/or minimization of risk.

An interesting finding was that maternal anxiety promoted accuracy in the current study. Mothers with less anxiety were found to underestimate their child's risk, both initially and across time. This finding compliments previous findings that mothers who are less anxious over time are more likely to underestimate their newborn's risk (Carmichael et al., 2003).

Supported hypothesis 2. Mothers from ethnic minority groups and those with lower levels of education will have less accurate risk perceptions. In this sample maternal educational level was found to be significantly related to maternal accuracy at

Follow-Interview 1. Maternal African American ethnicity was associated with maternal underestimation of infant risk over time. Preliminary work with this population had found maternal accuracy to be associated with a higher degree of maternal education, ethnic minority status, a lower degree of newborn risk, and less time since notification of newborn risk status (Carmichael et al., 2003). In general, in the genetic screening literature, woman from ethnic minority groups and woman with lower levels of education have been reported to retain less information about screening programs (Browner et al., 1996; Donovan & Tucker, 2000; Hughes et al., 1997).

Supported hypotheses 3. Mothers scoring high in information seeking will have more accurate risk perceptions, both initially and across time. Both objective measures of maternal information-seeking (e.g., length of notification phone call, requesting general PANDA information) and self report of information-seeking were found to be related to maternal accuracy or underestimation of risk. It is likely that information seeking mothers cope more effectively with stressful situations by using more information-obtaining coping strategies rather than avoiding the situation (Scheier, Weintraub, & Carver, 1986; Carver, Scheier, & Weintraub, 1989). This is consistent with the monitoring and blunting hypotheses, which postulates that information facilitates coping for high monitors, or information seekers, and this effect has been reported in the genetic screening literature (e.g., Schwartz, Lerman, Miller, Daly, & Masny, 1995).

Supported hypothesis 4. Tailored risk information letters reiterating what a mother was told over the phone regarding the child's risk will facilitate more accurate recall of the child's risk status compared to a no letter condition. Tailored print interventions reiterating risk have been found to facilitate more accurate recall of risk status compared to a no letter condition (Evans et al., 1993, Garrud et al., 2001, Rothert et al., 1997). This effect was found to be stronger over time, perhaps allowing mothers' time to reflect and review their child's risk and consult with medical professionals, friends, or family members. Most frequently in the literature, the reported effect of tailored risk print interventions has only been related to adherence to behavioral recommendations, with little examination of the mediating factor of perceived risk. Further investigation in this area is warranted to best determine not only the impact of tailored print interventions on adherence but also on the mediating variables of risk perceptions and anxiety.

Unsupported hypothesis. Having a family history of diabetes of any type will be associated with increased accuracy of risk perception. The findings in the literature are mixed with respect to this hypothesis, which was based on preliminary findings in the PANDA population. Several factors could explain the differences found between the current study population and a prior study which found an effect for family history on accuracy (Carmichael et al., 2003). One consideration is the sample makeup. Specifically, there are significant differences in the rates of newborn risk categories between the present study and that of Carmichael et al. (2003) and consequently, family rates of diabetes. Our current study, with an N of 190, placed 70% of newborns in the moderate risk category, with the remainder in the high risk category. There was only one extremely high risk infant. In contrast, Carmichael et al. (2003) reported that their sample of 351 infants consisted of 60% at moderate risk, with 34.7% at high risk and 5.3% at extremely high risk. This made for a larger sample of relatives with a family history of diabetes and in particular, with a first degree relative with Type 1 diabetes. In addition, the smaller sample size of the current study may have had insufficient statistical power to detect the effects of these variables in the present sample.

Maternal Anxiety in Response to Risk Notification

The results of the analysis supported the following hypotheses 1)Anxiety will be initially significantly elevated, 2) Over time, maternal anxiety levels will significantly decline to normal levels; 3) Mothers from ethnic minority groups will report greater anxiety after being notified of the child's increased risk, 4) Higher actual infant risk will be related to higher levels of maternal anxiety, 5) Mothers scoring higher in optimism will report lower levels of maternal anxiety than mothers lower in dispositional optimism, and 6) Mothers scoring higher in information seeking will report higher levels of anxiety than mothers scoring lower in information seeking.

The data did not support the following hypotheses 1) Mothers with lower levels of education will report greater anxiety after being notified of the child's increased risk; 2) Mothers who state satisfaction with the amount of information they receive will report less anxiety than those who were not satisfied, 3) The amount of information preferred and the amount of information obtained will interact, such that high levels of information seeking with low levels of satisfaction will result in high levels of anxiety, or 4) Mothers whose children have a family history of diabetes will report more anxiety than mothers whose children have no family history of diabetes

Supported hypotheses 1 through 4 Anxiety will be initially significantly elevated for some sub-groups of mothers Over time maternal anxiety levels will decline to normal levels Mothers from ethnic minority groups will report greater anxiety after being notified of the child's increased risk. Higher actual infant risk will be related to *higher levels of maternal anxiety* The second major conclusion of the current study is that mothers of newborns at-risk for Type 1 diabetes experience an elevation in anxiety upon notification of risk and a decrease to normative levels approximately three months later In this study, African-American mothers and mothers of high risk infants reported significant elevations in their anxiety levels shortly after the time of risk notification. This is consistent with prior studies on the maternal response to newborn at-risk status for Type 1 diabetes (e.g , Johnson et al , in press), which found elevations in anxiety for mothers of high risk infants and Hispanic mothers These findings are also consistent with other reports of anxiety related to genetic risk communication (e g., Bekker et al, 1994, Watson et al., 1992) in terms of similar (e g., Tercyak & Johnson, 1995) and unrelated at-risk populations. In general, however, anxiety levels for the sample as a whole were not clinically elevated

Across time African-American ethnicity was not found to be related to the maintenance of anxiety Maternal African-American identification and Follow-Up Interview 1 Anxiety were significantly correlated (r=.309, p= 000), making the effect of ethnicity weaker when initial anxiety is being controlled.

Supported hypothesis 5 Mothers scoring high in optimism will report lower levels of maternal anxiety than mothers lower in dispositional optimism For the mothers of newborns at risk for diabetes in the current study higher levels of dispositional optimism were related to lower anxiety both initially and over time This is consistent with previous findings that mothers who hope that the risk status will "magically" disappear tend to maintain their anxiety time (Johnson, 2001, Johnson & Carmichael, 2000) Higher levels of optimism have been consistently reported to be related to lower levels of distress due to minimization of risk or avoidance coping (Clark, 2000; Michie, Bobrow, & Marteau, 2001). In the present study, mothers who reported themselves to think in more optimistic, half-glass-full terms may have "reframed" the concept of their child's risk. Despite maintaining an accurate risk perception, the optimistic mothers also maintained lower anxiety levels than those mothers who reported lower levels of dispositional optimism. This leads one to wonder whether this effect is due to more passive avoidance coping or more "positive" coping styles such as problem focused coping or cognitive reframing.

Supported hypothesis 6. Mothers scoring higher in information-seeking will report higher levels of anxiety than mothers scoring lower in information-seeking. It was expected that mothers scoring higher in information-seeking would report higher levels of anxiety both after notification and over time, based on the findings of Lerman et al. (1996) who reported that monitors had higher distress in genetic testing situations due to probable higher perceived risk and more intrusive thoughts. Our findings indicate that in fact, maternal information-seeking does lead to increased anxiety and that this effect remains strong over time. This finding is likely to remain consistent over time, particularly as more information seeking will lead to the knowledge that there is nothing that can be done preventatively in this situation.

Unsupported hypotheses 1. Mothers with lower levels of education will report greater anxiety after being notified of the child's increased risk. This finding was reported by Johnson et al. (in press) in a prior PANDA sample. In this study, maternal education was found to be a significantly predictor of initial anxiety, but the effect was lost when African American ethnicity was added to the regression model. It is possible that education is mediating the effect of ethnicity on anxiety for this sample and/or our smaller sample size is limiting the expression of the effect in the current study

Unsupported hypotheses 2 and 3 Mothers who stated satisfaction with the amount of information they received will report less anxiety than those who were not satisfied and *The amount of information preferred and the amount of information obtained will interact*, such that high levels of information seeking with low levels of satisfaction result in high levels of anxiety No studies to date have examined satisfaction with information received and anxiety in a genetic testing situation, and we found no relationship on anxiety on whether or not a mother reported satisfaction with the amount of information received As such, no such conclusions can be drawn from this population perhaps due to the limited measurement of "information-seeking" (which some measure as a monitoring coping strategy instead of assessing actual behaviors) and "satisfaction "

Unsupported hypothesis 4 Mothers whose children have a family history of diabetes will report more anxiety than mothers whose children have no family history of diabetes. Again, this effect was hypothesized based on the findings in a prior PANDA sample (Johnson et al., in press) The prior study found that over time, mothers whose infants had a first degree diabetic relative tended to remain more anxious Given that our sample only contained nine infants with a first degree relative with diabetes, our sample size and composition limited findings related to this hypothesis.

Maternal Intent to Continue with PANDA Study

The final area of investigation of the current study was maternal intent to continue with the PANDA study Results of data analysis found support for the following hypotheses 1) Mothers with children who have a family history of diabetes will be more likely to report intentions to continue in PANDA than mothers of children with no family history of diabetes, 2) Mothers receiving a tailored risk information letter post-notification will be more likely to report intentions to continue in PANDA than those mothers who did not receive a tailored risk information letter; 3) Mothers who report and underestimation of their child's risk will be less likely to report intentions to continue in PANDA, and 4) Over time, the number of mothers willing to continue in PANDA will decline.

Hypotheses which did not find support from the data include: 1) Caucasians will report greater intentions to continue in PANDA than African-Americans, 2) Mothers of higher risk infants will report greater intentions to continue in PANDA than mothers of lower risk infants, 3) More anxious mothers will report intentions to continue in PANDA, 4) Mothers high in dispositional optimism will be less likely to report intentions to continue in PANDA, and 5) Mothers with greater information-seeking behaviors will be more likely to report intentions to continue in PANDA.

Supported hypothesis 1. Mothers with children who have a family history of diabetes will be more likely to report intentions to continue in PANDA than mothers of children with no family history of diabetes. The factors associated with intention to continue in the study included initial intent at the notification phone call and having any family history of diabetes. In previous studies (i.e., Johnson, 2001), newborn risk status and initial anxiety were factors that predicted intent to continue with the study. Lerman et al. (1990) reported that a positive family history of cancer predicted adherence to health screenings in women. In addition, one would also expect populations with a family history of disease to be at higher risk and thus, be more strongly targeted for participation.

Supported hypothesis 2 Mothers receiving a tailored risk information letter post-notification will be more likely to report intentions to continue in PANDA than those mothers who did not receive a tailored risk information letter In particular this researcher felt that an effect might be observed between letter conditions, so that mothers receiving a rationale for participation (altruistic or monetary) may be more likely to participate than a mother simply receiving reiteration of infant risk There was no effect found for type of letter based on literature that written reminders or follow-up (that presumably provided a rationale for participation) increased adherence to pap screening and immunizations (e g , Kash et al, 1992, Szilagyi et al , 2000)

However, the current study found an effect on intentions to continue in the study for those receiving any type of tailored risk information letter Mothers who simply received a tailored risk information letter, either with or without a monetary or altruistic slant, were more likely to state intentions to participate in PANDA More recent studies (Clark et al. 2002 Rimer et al, 2001, Sweet, Willis, Ashida, & Westman, 2003, Valanis et al, 2003) similarly report positive effects for tailored risk communication messages that are in print, but indicate that additional reinforcement measures, in particular, follow-up telephone counseling or physician calls (Lipkus, Rimer, Halabi & Surgio 2000) may be more effective in both increasing motivation for action/behavior changes

Supported hypothesis 3 Mothers reporting an underestimate of their child s risk will be less likely to report intentions to continue in PANDA The data indicated that mothers who were less likely to report an underestimate of their infant s risk were more likely to report intentions to continue in PANDA, in other words, those mothers who minimize risk do not show interest in further study participation. This is consistent with literature that has reported that those who perceive themselves to be at a high risk for a disease tend show interest in testing, while those who minimize or dismiss risk indicate no interest (Cameron & Diefenbach 2001, Codori et al., 1999, Croyle & Lerman, 1995 Kash et al , 2000; Lerman, Daly, et al., 1994, Lerman, Rimer et al., 1994 Marteau et al , 1991, 1992) Johnson et al (1990) reported that those persons at high risk for developing Type 1 diabetes tended to minimize their risk for disease development. Disease-specific anxiety as well as having a family history of disease (e.g , Watson et al. 1992) may mediate this relationship. Lerman, Schwartz et al (1997) note, it makes sense that distress promotes risk assessment in the cancer screening literature (e.g., Lerman, Miller et al , 1991), as knowing one is at-risk affords you the opportunity to either reduce uncertainty, minimize distress, or provide you with the opportunity to consider methods of prevention rather than simply adhering to disease detection efforts.

Supported hypothesis 4 Over time more mothers were less likely to report intentions to continue with PANDA. This is consistent with previous findings described previously within our laboratory with a preliminary population of mothers with infants at-risk for the development of Type 1 diabetes (unpublished data)

Unsupported hypothesis 1 Caucasians will report greater intentions to continue in PANDA than African-Americans We found, as hypothesized, a relationship between ethnicity and intentions to continue although our findings were contrary to expectations Analyses indicated that Caucasian mothers were less likely to state intentions to participate in PANDA over time This finding is surprising as in general the literature indicates that African-Americans may be more reluctant to participate in research (Jones, 1983, Markel 1992) or even in genetic counseling programs (Kuppermann et al , 1996, Lerman et al., 1999) Lower levels of uptake are often associated with limited education and awareness, or ethnocultural, transportation, language, or other barriers. In the present sample, there was an correlation between Non-Caucasian status as a whole to educational or income measurements, indicating that socioeconomic barriers may not be the mediating factors for the present population. However, intent to participate in genetic studies and actual participation rates are often highly discrepant, perhaps indicating a positive response bias on the part of ethnic minority mothers in this sample

Unsupported hypothesis 2 Mothers of higher risk infants will report greater intentions to continue in PANDA than mothers of lower risk infants This hypothesis was based on preliminary findings with a population of PANDA mothers which found that having an infant at a higher risk was related to whether or not a mother appeared for further PANDA studies The difference in outcome measurement between the two studies may explain these findings as the current study is measuring maternal intent to participate while the previous findings were related to actual maternal study participation behavior

Unsupported hypothesis 3 More anxious mothers will report intentions to continue in PANDA. Mothers with higher anxiety levels have been shown to be more likely to continue with PANDA participation (Carmichael et al , 1999) Much other literature regarding screening and behavioral outcomes has demonstrated that disease-specific worry is related to intentions to use genetic testing (e g. Cameron & Diefenbach, 2001), Croyle & Lerman, 1993 Lerman et al , 1994), but it is unclear whether this is a linear or curvilinear relationship between anxiety and action to reduce a health threat (e.g., Kash, 1992). Again, we may be missing the effect as the current study assessed maternal intent while previous findings are related to actual maternal behavior. As found previously in the literature, high rates of interest do not necessarily lead to high rates of utilization, and this discrepancies between attitudes and behavior is deserved of further examination (Markel et al., 1987, Meissen & Berchek, 1987, Quaid et al., 1989).

Unsupported hypothesis 4. Mothers high in dispositional optimism will be less likely to report intentions to continue in PANDA. In the present study, dispositional optimism was not found to be related to stated intentions to continue in the study. As found previously, dispositional optimism was noted to be related to lower anxiety levels, likely due to decreased perception of risk/harm associated with an optimistic outlook. In two of the only studies to examine this concept to date, Biesecker et al. (2000) and Bosompra et al. (2000) found that those individuals high in optimism were less likely to participate in genetic screening than individuals scoring low in optimism. As anxiety does not appear to be influencing maternal intentions to participate in PANDA, it is not surprising that no effect was found for optimism. Again, this is believed to be due to the measurement of maternal intentions to participate instead of actual participation rates.

Unsupported hypothesis 5. Mothers with greater information seeking behaviors will be more likely to report intentions to continue in PANDA. Mothers who were either self-reported or observed "information-seekers" were believed to be more likely to participate in further testing. Participation in research studies may be viewed as an additional form of information seeking about health status. Monitors (information seekers) have been found to be more adherent to behavioral recommendations such as attending cancer screenings (Wardel et al. 1995). It is possible that effect was not large enough to detect in this sample due to sampling bias; those mothers responding to initial risk result letters and those who agree to participate in our interviews may be more likely in general to participate in PANDA than may have been seen if a more representative sample were available.

Prior data obtained from a population of mothers who had newborns identified as at-risk for diabetes included not only maternal intent to continue but measured actual participation in further PANDA studies, unlike the present sample. As mentioned previously, it is likely that a potential discrepancy exists between maternal intentions of continuing in PANDA and the actuality of having one's child participate in more anxiety-provoking aspects of the study such as an invasive and potentially painful procedures (i.e., child blood draws). This illustrates the difficulty in assessing intent because it is based solely on the mother's report and does not consider other factors. If the goal is to retain as many mothers as possible for future assessments and child monitoring, it would be worthwhile to develop a protocol to predict mothers who are at risk for not continuing with a study and to develop effective educational and/or behavioral interventions for the purpose of increasing the likelihood of maintaining study enrollment.

Limitations of the Current Study

Attrition. In many longitudinal studies, attrition is a barrier to full understanding of the topic of investigation. In the current study, the participation rate for the Follow-Up Interview 2 was consistent with prior PANDA psychological studies (Carmichael et al. 2003, Johnson et al., in press). Of interest to both biomedical and psychosocial researchers are the factors that prompt mothers to not continue with the study, whether a lack of interest in the study, a minimization of risk that may already seem low, too high or too low anxiety levels, especially if there is no family history of diabetes and diabetes itself may seem unfamiliar, or because of other unknown or unrelated circumstances. It has been observed that the responsibility falls on the researchers in this area to identify mechanisms to retain individuals at-risk for not continuing with studies and to identify mechanisms whereby such individuals may be prompted to increase risk information accuracy, cope more adaptively, and maintain an interest in study participation. The results of the current study certainly suggest that family history of disease, ethnicity, and the development of a tailored risk information print materials are relevant variables to consider when developing specific protocols to increase the likelihood of staying in a longitudinal study.

Lack of Control Group. One of the greatest limitations of the current study is the lack of a control group. Such a control group would have allowed for expanded generalizability of findings as it would have allowed comparison between mothers of at-risk infants and mothers of non at-risk infants who were notified that their newborns were not at-risk for Type 1 diabetes through the PANDA study. A control group to compare levels of anxiety and accuracy would have proved beneficial when considering the outcomes of the study, but are often not provided in study design when examining predictive genetic testing. Typically, researchers in this area focus on first establishing genetic risk through testing and then provision of long-term monitoring. Economic constraints typically limit the number of individuals studied to those typically at highest risk for the disease. Unfortunately, psychological studies linked to genetic testing protocols are often implemented at a later time, after which it becomes difficult to manipulate study groups Nonetheless efforts should be made to incorporate control groups into predictive genetic testing programs

Strengths of the Current Study and Final Comments

To date there are few published studies documenting the psychological impact of predictive tests for Type 1 diabetes (Carmichael et al , 2003, Johnson & Carmichael, 2000, Johnson & Tercyak, 1995, Johnson et al., in press, Johnson et al , 1990, Roth et al , 1994, Roth et al , 1996) The current study addressed the specific questions of whether mothers understand the information they received regarding their newborns risk for Type 1 diabetes development, the impact of the method of information receipt, maternal reactions to the news, and the resulting effect on intent to continue with the PANDA study It is one of the few studies to focus on parental interpretations of risk in relation to medical data and their child and the only one so far to implement an intervention in an attempt to modify risk perceptions and effect study participation in this population.

One strength of this study is that it demonstrates that a simple low-cost intervention, a tailored risk information letter, appears to increase maternal understanding of infant risk over time This intervention also promotes maternal intent to participate in the study at no expense to maternal anxiety The fact that this study investigates how parents understand risk in relation to their children also contributes to the limited literature that exists in this area. Our findings of fairly high levels of accurate understanding are encouraging In addition, this study also finds strength in the replication of main themes related to past findings with PANDA mothers Namely, mothers have a fairly good understanding of infant risk, which tends to lean towards minimization/underestimation over time related to maternal demographic and study variables. Second, mothers experience elevated levels of anxiety upon notification of the newborn's at-risk status. That level of anxiety is of normal levels and decreases over time. In addition, others have pointed out, no significant increase in anxiety when provided with detailed, anxiety-provoking information may in itself be a positive outcome (Garrud, Wood, & Stainsby, 2001).

Making the decision to participate in genetic screening studies is not easy and may be influenced by a variety of factors, including risk perception, education and ethnicity, previous experience with diabetes, information-seeking style, dispositional optimism, and anxiety. In this population, no intervention to increase accuracy of risk perception subsequent to newborn genetic screening had been tested. Similarly, the role of dispositional optimism, information-seeking style, and satisfaction with information obtained had not yet been examined as potential predictors of risk perception, anxiety, or stated intentions to participate in a longitudinal study. It will be important to determine further what type of risk notification strategies work best to ensure that mothers feel satisfied with the information they receive, and are well-informed about their decision to continue or decline participation in a longitudinal study of their child. The current study has promoted more specific understanding of the psychological and family factors related to risk information receipt, anxiety, and maternal intent to participate in studies of newborn genetic screening for Type 1 diabetes.

APPENDIX A
DIABETES RISK FACTORS

DR/DQ alleles/genotypes	With First Degree Relative	Without First Degree Relative	% of in general population	% of those who have Type I
DR 3/4, DR 4/4 DQ 0201/0300 DQ 0300/0300	20-25/100 (1/4-5) **Extremely High Risk**	5/100 (1/15) **High Risk**	5%	>50%
DR 3/4, DR 4/X DQ 0201/0201 DQ 0300/X (X not= 0602)	10/100 (1/10-15) **High Risk**	2/100 (1/45-60) **Moderate Risk**	10%	30-40%
DR 3/4 or X/X DQ X/X	1/125 Intermediate Risk	1/600 Very Low Risk	85%	10%
DR 3/4 or X/X DQ 0602	1/15,000 Protective	1/15,000 Protective		

Note. X is a non-defined allele 75% of the time X= DR 4 or DQ 0301

APPENDIX B
PROCEDURAL FLOW OF STUDY

INITIAL GENETIC SCREENING OF NEWBORN FOR DIABETES RISK AT BIRTH (PANDA STAFF)

↓

SCRIPTED NOTIFICATION OF RESULTS TO MOTHER BY PANDA STAFF MEMBER

↓

RISK INFORMATION LETTERS RANDOMIZED AND MAILED TO MOTHERS AFTER NOTIFICATION

↓

STRUCTERED TELEPHONE INTERVIEW WITH MOTHER ~30 DAYS POST-NOTIFICATION (FOLLOW-UP INTERVIEW 1)

↓

SECOND STRUCTURED TELEPHONE INTERVIEW WITH MOTHER ~3.5 MONTHS AFTER FOLLOW-UP INTERVIEW 1 (FOLLOW-UP INTERVIEW 2)

↓

OPPORTUNITY TO PARTICIPATE IN PANDA PART II BLOOD DRAW

APPENDIX C
SAMPLE RISK NOTIFICATION INTERVIEW

High Risk

With First Degree Relative

____DQ201/0201 ____DQ 0300/X (Xnot=602)

Hi (mom's name)____________ This is ___________________(study coordinator) from the University of Florida Diabetes Research Office. Is this a good time to talk? As you know, we now have the results of a study that (child's name)_____________ participated in on (date)_____________, called the PANDA study. As you might recall, we tested your baby's blood. We wanted to see if your baby/child had any genes that might put him/her at risk for developing type I diabetes.

We have found that your baby/child is at a **high risk** for developing diabetes. Now, please keep in mind that this does NOT mean that your child will definitely get diabetes. It just means that out of 100 children with the same genetic markers as your child, and who also have a parent or sibling with diabetes, there would be about 10 babies that would go on to develop diabetes. Now that you have this information, you may ask "what now?"

There is a second part of our study, where we would like to follow your child at 6 month intervals. We want to learn why some children with your child's genes go on to develop diabetes and why other children with the same genes never develop diabetes. If you agree to participate in Part II of our study, we will call you to ask you general questions about your child's health and development. When your child is at least 6 months of age, we would perform a second blood test. In this test, we draw blood from the arm and screen for the presence of autoantibodies, which are more markers for the disease. If the results are negative, we will continue to screen your child every six months. If the test results are positive, then we would like to follow your child a little more closely and discuss the meaning of having a positive antibody result with you at that time.

Each time we test your child's blood, you will receive a $20 reimbursement for your time, unless you are an employee of the state of Florida. We want you to know that your child is very special. Your child's participation in the study can provide extremely important information that may help us find a way to prevent diabetes.

Now I've just given you this information, do you have any questions for me about what I've just told you, or any other questions about diabetes and the study we would like to continue to do? (For Coordinator: If yes, see below. If no, obtain decision.)

Check all items discussed and indicated who initiated the topic.

What does PANDA mean? ____Parent _____coordinator

Prospective Assessment in Newborns for Diabetes Auto-immunity

How did you find out that my child is at high risk? What did you look at? ____Parent _____coordinator

Well, each child receives one gene from their mother and one gene from their father. Each carries a risk for developing diabetes. The combination of the genes is what determines risk.

What percentage of the children tested are at increased risk, and in particular, how many are in the high risk category? ____Parent _____coordinator

Approximately 26 out of every100 children that we have screened in our study have been found to be at a higher than average risk for developing type I diabetes. In our study, approximately 4% of the children have been found to be at the same high-risk level for developing Type I diabetes as your child. Like your child, these children do have a parent or sibling with Type I diabetes.

What percentage of the children in the general population has these high risk genetic markers? ____Parent _____coordinator

In the general population, about 10 out of 100 children, or 10%, have the same genetic markers as your child.

How many children with diabetes have this gene? ____Parent _____coordinator

Approximately 30-40% of all people with diabetes have the same genetic markers as your child.

What is the general population risk for developing diabetes? ____Parent _____coordinator

Approximately 1 out of 300 people in the general population has Type I diabetes.

What will you be looking for exactly in the second part of the study? ____Parent _____coordinator

We will be looking for biological markers of diabetes in the blood. There are certain antibodies that are more likely to be found in the blood of people who develop Type I diabetes, such as antibodies to islet cells (cells in the pancreas that produce insulin), and insulin, along with other proteins. These "markers" may be present in the blood years before the disease begins.

How accurately do these markers predict disease development? ____Parent _____coordinator

Approximately 20-50% of people with more than one marker go on to develop diabetes.

Where/how/how much/how often will be child's blood be drawn? ____Parent _____coordinator

We would like to test your child's blood beginning at 6 months of age. We can draw either a few tablespoons from the arm or take a very small sample called a "bullet" from the heel. Most children do very well with the blood draw and our nurse is very experienced at drawing blood from children. We would like to test your child's blood approximately every 6 months. We will let you know when it is time for a blood test.

Are there any risks of participating in this study? ____Parent _____coordinator

You and/or you child may experience distress during the blood draw or some minor bruising could occur There also may be psychological affects for example you might experience some anxiety or depression at the news if your child is found to be at increased risk of developing diabetes. You may have to decide whether this information is something you want to share with the rest of your family, your doctor, or whether or not you want to have other members of your family tested

Will it cost me anything? ____Parent _____coordinator

Other than the gas and time it may take to either come to the Clinical Research Center at Shands Hospital, or to one of our participating labs, or your pediatrician, it will not financially cost you anything. You will be compensated $20 per blood draw for any costs you might incur

What happens if my child has these antibodies? ____Parent _____coordinator

The biological makers, or antibodies indicate some important changes in the body However they do not necessarily mean that your child will go on to develop diabetes. If there are markers present, the research team will want to follow your child more often They may want to test your child s blood every 3 months Sometimes autoantibodies appear and then may disappear for reasons we don't yet understand

How accurate are your tests? ____Parent _____coordinator

The tests of your child s genes are accurate 99% of the time

What kinds of diabetes are you looking for? What are the signs and symptoms? ____Parent _____coordinator

We are studying Type I diabetes. Diabetes is an autoimmune disease, which means that for some reason the body starts to destroy it's own cells that make insulin. Insulin is the hormone that helps to move sugar from the blood stream into the body s cells where it can be used for energy We are not sure what causes it exactly It seems to be a combination of environmental and genetic factors and that's what we are trying to figure out Signs and symptoms that may indicate diabetes include fatigue, increased hunger weight loss, increased thirst and high amounts of urination

What is the difference between Type I and Type II diabetes? ____Parent _____coordinator

Type I is also known as insulin-dependent or juvenile-onset diabetes and typically happens in childhood or adolescence It is treated by injecting insulin and monitoring of blood sugar This is the type of diabetes that we are studying Type II diabetes typically occurs as people get older It is often found in older people who are overweight or have a high degree of body fat It is treated through diet and exercise insulin pills, and sometimes insulin injections

Is there any cure? What can I do? ____Parent _____coordinator

At this time, there is not a cure for diabetes However the disease can be treated with insulin, exercise, and diet. If your child is at least 3 years of age, there is a study available called the Diabetes Prevention Trial (DPT) which looks at the use of insulin to see if that helps to slow down or stop the progression of diabetes in at risk people For now there is nothing else you can do to prevent diabetes We encourage all parents to teach good health habits such as proper diet and exercise and have your child monitored closely

Should my child avoid all sugar/sweets? ____Parent _____coordinator

At this time, there is no evidence that eating sugar "causes" diabetes. We recommend a normal, well-balanced diet

Who will have access to this information? ____Parent _____coordinator

We keep all information confidential and will only release information about your child if you should request However researchers directly involved with the study will have access to your child s results, but your child s name and medical record number will not be used

Should I tell my doctor about this? ____Parent _____coordinator

That is up to you whether you d like your doctor to know about your child's test results. However once this information is recorded in your child s doctor s medical records, insurance companies could obtain these results or the insurance company might try to use the information to avoid coverage if your child develops diabetes Because testing is not considered a standard clinical test, it is not currently legal for insurance companies to do this.

Can I have my other children tested for their risk? ____Parent _____coordinator

Certainly we test all your children, as well as you and your spouse, to determine diabetes risk

What will happen to any extra blood samples you may have? ____Parent _____coordinator

Only if you agree to it at the time your child's blood is drawn, the blood samples may be stored for additional genetic testing to learn more about diabetes This information may or may not benefit you directly Every attempt will be made to keep all information confidential

If you have any new developments in diagnostic technology, would we be notified if our risk status changes? ____Parent _____coordinator

If we discover new information that may be important to your or your child s personal health and risk for developing diabetes, we would certainly let you know this new information

Can you send me more information about (check one/both) ____Parent _____coordinator

_______the PANDA study and/or ____diabetes.

Information sent

_____letter sent with specific information the parent requested_______________________
_____diabetes information leaflet and/or PANDA Part II information
_____other______________________________

Note outcome:

___mom declined participation
___mom deciding
___mom wants to participate, will schedule later
___mom wants to participate, scheduled for (date) __________ at (place)____________
___Mailout sent for ____child ____mother ____father ____siblings

Length of call in minutes ______

Who initiated the call ______Parent in response to letter
______ Parent called, without letter having been sent
______Coordinator called after no response from letter

APPENDIX D
FOLLOW UP LETTER WITH MONETARY EMPHASIS

November 10, 2003

Mrs. Jane Smith
1234 Archer Road
Gainesville, FL 32608

Dear Jane,

Thank you for Connor's participation in Part I of our PANDA study (Prospective Assessment in Newborns for Diabetes Auto-Immunity) This letter is being sent out to all study participants, regardless of your family's decision to participate in Part II of PANDA at this time. As I explained to you today over the phone, we place children into certain "risk groups" based on their genetic test results and family history of diabetes. Connor was found to be at High Risk for developing Type I diabetes This means that out of 100 children with the same genetic markers as Connor, and no parent or sibling with diabetes, 5 children would be expected to go on to develop Type I diabetes. As we discussed, this does not necessarily mean that Connor will go on to develop Type I diabetes

In addition to the genetic screening, we do offer a second part of the study for "at risk" children We look for markers in the blood, called autoantibodies. These markers indicate that the process causing diabetes may have started If you are willing, we would like to continue to test Connor's blood once each year to see whether this process of diabetes development has begun If you have already communicated your decision to us, thank you. If you have further questions or concerns, or would like to confirm an appointment, please do not hesitate to contact us You may call us on the toll free number 1-800-749-7424, ext 2-7836

We can offer you $20 for each blood test as compensation for your time and expense for your participation (unless you are a State of Florida employee)

Thank you for your participation in Part I of PANDA we welcome working with your family in Part II

Sincerely,

Diabetes Research Coordinator
The PANDA Study

APPENDIX E
FOLLOW UP LETTER WITH ALTRUISTIC PLEA

November 10 2003

Mrs Jane Smith
1234 Archer Road
Gainesville, FL 32608

Dear Jane,

Thank you for Connor's participation in Part I of our PANDA study (Prospective Assessment in Newborns for Diabetes Auto-Immunity) This letter is being sent out to all study participants, regardless of your family's decision to participate in Part II of PANDA at this time. As I explained to you today over the phone, we place children into certain "risk groups" based on their genetic test results and family history of diabetes. Connor was found to be at High Risk for developing Type I diabetes. This means that out of 100 children with the same genetic markers as Connor and no parent or sibling with diabetes, 5 children would be expected to go on to develop Type I diabetes. As we discussed, this does not necessarily mean that Connor will go on to develop Type I diabetes

In addition to the genetic screening, we do offer a second part of the study for "at risk" children We look for markers in the blood, called autoantibodies. These markers indicate that the process causing diabetes may have started If you are willing, we would like to continue to test Connor's blood once each year to see whether this process of diabetes development has begun If you have already communicated your decision to us, thank you. If you have further questions or concerns, or would like to confirm an appointment, please do not hesitate to contact us You may call us on the toll free number 1 800-749-7424, ext. 2-7836.

Your child is extremely special   His/her participation in the study may help us find a way to prevent diabetes

Thank you for your participation in Part I of PANDA, we welcome working with your family in Part II

Sincerely

Diabetes Research Coordinator
The PANDA Study

APPENDIX F
PHONE INTERVIEW SCRIPT FOLLOW-UP INTERVIEW 1

Mother's Name ______________________ **Phone** ________________________

Child. ___________________________ **Date:**___________________________

Risk Status _____________________________with/without first degree relative (*circle one)*

PANDA Study Interview #1

Screening of newborns/children to identify subjects at high risk for developing insulin-dependent diabetes, Part III Maternal perceptions, reactions, and experiences (IRB# 353-95)

Hello, my name is ________________ *and I'm calling from the UF Diabetes Research Office I understand that you spoke with Karrie North last week regarding your baby's genetic test results. I'd like to ask you a few questions about your understanding of your child's participation in this study. Do you have a few minutes to speak with me right now? In the event that you feel uncomfortable answering any question, you are free to skip that question. Please keep in mind that all information is kept confidential.*

Can you tell me, from what Karrie told you, what is your understanding about your child's risk for developing diabetes?

SPONTANEOUS

__very very low risk	___1/15 000	___moderate risk	____2/100	__don't know
__low risk	___1/600	___high risk	____5/100 or ____10/100	
__intermediate risk	___1/25	___very high risk	____1/5 or ____20-25/100	

If both a number and a category were not mentioned, or if any one was wrong, PROMPT
Do you remember any numbers or categories?

___very very low risk	___1/15,000	___moderate risk	____2/100	__don't know
__low risk	___1/600	___high risk	____5/100 or ____10/100	
__intermediate risk	___1/25	___very high risk	____1/5 or ____20-25/100	

(If both were given correctly skip this question If the mother gave an incorrect answer did not give two answers, or said she did not know give her the following recognition task Read all categories and numbers) Kerrie might have told you that your child was more or less at-risk for developing diabetes than other people in the general population I m going to read some categories and give you some numbers and I want you to tell me after each one, whether or not it was the risk category that Kerrie told you your baby was in Okay? Were you told your baby was at

1 Very very low risk (~1/15 000) **yes/no**
2 Low risk (~1/600) **yes/no**
3 Slightly higher risk (~1/125) **yes/no**
4 Moderate risk (~2/100) **yes/no**
5 High risk (~5 or 10/100) **yes/no**
6 Extremely high risk (~1/5) **yes/no**

I want you to think about your child's risk for developing diabetes. I'm going to describe some feelings to you, and I want you tell me how you feel right now at this moment.

GIVE STAI-SF
Would you say you currently feel

Not at all calm	*somewhat calm*	*moderately calm*	*very calm*
Not at all worried	*somewhat worried*	*moderately worried*	*very worried*
Not at all relaxed	*somewhat relaxed*	*moderately relaxed*	*very relaxed*
Not at all tense	*somewhat tense*	*moderately tense*	*very tense*
Not at at-ease	*somewhat at-ease*	*moderately at-ease*	*very at-ease*
Not at all nervous	*somewhat nervous*	*moderately nervous*	*very nervous*

Does anyone else in your family have diabetes? *(include information regarding the participant s estimating of diabetic control and how problematic the disease has been for the patient and family members)*

Relationship to child (circle)	Type	Age at onset	Live with you? 1=yes 2=no	Duration	Control*	Problems**
Mother						
Father						
Full Sibling						
Full Sibling						
Half Sibling						
m grandmother						
m grandfather						
m grt grandmother						
m-grt grandfather						
p-grandmother						

p-grandfather						
p-grt.grandmother						
p-grt grandfather						
(other)						

Note Indicate if they DON'T KNOW *1=excellent 2=good 3=fair 4=poor 5=DK **1=none 2=a few 3=several 4=a lot 5=DK

Please indicate how much you agree or disagree with the following statements. There aren't any correct answers, so just answer according to your own general feelings rather than how you think others might answer (First ask if they agree disagree or are neutral about the question Then distinguish between strongly agree/disagree and agree/disagree)

SA=Strongly Agree A=Agree N=Neutral D=Disagree SD=Strongly Disagree

For example (1)I enjoy my friends a lot (2)It's easy for me to relax.

In uncertain times, I usually expect the best	SA	A	N	D SD
If something can go wrong for me it will	SA	A	N	D SD
I always look on the bright side of things	SA	A	N	D SD
I'm always optimistic about my future.	SA	A	N	D SD
I hardly ever expect things to go my way	SA	A	N	D SD
Things never work out the way I want them to	SA	A	N	D SD
I'm a believer in the idea that "every cloud has a silver lining."	SA	A	N	D SD
I rarely count on good things happening to me	SA	A	N	D SD
Overall, I expect more good things to happen to me than bad	SA	A	N	D SD

Since Kerrie talked to you about your child s risk, did you

(1)speak with a family member to ask about your child s family history of diabetes? Y/N
(2) talk to your pediatrician or other medical professional about your child's risk Y/N

(3) call study personnel to discuss your child's risk? Y/N

(4)read a book or other literature on diabetes or diabetes risk? Y/N

(5)search the Internet for information on your child's risk? Y/N

Satisfaction with information received.

Which of the following best describes the amount of information received about the

study

___You received too little information about the PANDA Part II study
___You received just the right amount of information about the PANDA Part II study
___You received too much information about the PANDA Part II study

At the present time what are your plans with continuing with the PANDA study and having your child's blood tested again in the future?

___No, you are not considering continued participating at this time

___You're not sure right now if you are going to continue participating in the PANDA study

___Yes, you are definitely going to continue participating in PANDA Part II (make sure they have 800 number)

What are your thoughts on having had your child tested at birth for diabetes risk?

(1)Glad you participated in the study
(2)Not sure about your participation in the study
(3)Wish you had not participated in the study

Moms DOB ______________
Mom's Race ___________
Child's Race ___________
1=Caucasian
2=African American
3=Hispanic
4=Asian
5=other
allow more than 1 to be selected

Parents Marital Status ______________
1=single parent, involved father
2=single parent, non-involved father
3=married
4=separated
5=divorced

Educational level of mother___________
1=some high school
2=graduated HS/GED
3=some college or trade school

Educational level of father ___________
4=graduated from college/trade school
5=some graduate school or professional
6=graduated from graduate school or professional program

# of children in the family ________(first child? Y/N)

Annual Total Family Income ___________

Thank you very much for your time tonight. If you have any questions about the study, please feel free to call Kerri North at 1-800-749-7424 ext. 27836, or locally at (352) 392-7836

APPENDIX G
PHONE INTERVIEW SCRIPT FOLLOW UP INTERVIEW 2

Mother's Name ____________________ **Phone** ______________________

Child ________________________ **Date** __________________________

Risk Status __________________________with/without first degree relative (*circle one)*

PANDA Study Interview #2

Screening of newborns/children to identify subjects at high risk for developing insulin-dependent diabetes, Part III Maternal perceptions, reactions and experiences (IRB# 353-95)

Hello my name is __________________ and I'm calling from the UF Diabetes Research Office. You might remember talking to be about __ weeks ago regarding your child's participation in the PANDA study I'd like to ask you a few additional questions about your child's participation in this study Regardless of whether or not you intend to have your child tested this is a separate interview about your feelings about this news Do you have a few minutes to speak with me right now?

Can you tell me, from what Korrie told you, what is your understanding about your child's risk for developing diabetes?

SPONTANEOUS

__very very low risk	___1/15,000	___moderate risk	____2/100	__don't know
__low risk	___1/100	___high risk	____5/100 or	____10/100
__intermediate risk	___1/25	___very high risk	____1/5 or	____20-25/100

If both a number and a category were not mentioned, PROMPT
Do you remember any numbers or categories?

___very very low risk	___1/15 000	___moderate risk	____2/100	__don't know
__low risk	___1/100	___high risk	____5/100 or	____10/100
__intermediate risk	___1/25	___very high risk	____1/5 or	____20-25/100

(If both were given, and were correct skip this question If the mother needs help or had at least one incorrect answer above give her the following recognition task. Read all categories and numbers) Kerrie might have told you that your child was more or less at risk for developing diabetes than other people in the general population I'm going to read some categories and give you some numbers, and I want you to tell me after each one whether or not it was the risk category that Kerrie told you your baby was in Okay? Were you told your baby was at

1 Very very low risk (~1/15 000) **yes/no**
2. Low risk (~1/600)**yes/no**
3 Slightly higher risk (~1/125) **yes/no**
4 Moderate risk (~2/100) **yes/no**
5 High risk (~5 or 10/100) **yes/no**
6 Extremely high risk (~1/5) **yes/no**

I'm going to describe some feelings to you I want you to think about how you feel right now at this moment, about your child s risk status for developing diabetes I know you answered these questions before when we first interviewed you, and your feelings may or may not have changed Just tell me how you presently feel about your child s risk for developing Type I diabetes

GIVE STAI-SF:
Would you say you currently feel

Not at all calm	*somewhat calm*	*moderately calm*	*very calm*
Not at all worried	*somewhat worried*	*moderately worried*	*very worried*
Not at all relaxed	*somewhat relaxed*	*moderately relaxed*	*very relaxed*
Not at all tense	*somewhat tense*	*moderately tense*	*very tense*
Not at at-ease	*somewhat at-ease*	*moderately at ease*	*very at-ease*
Not at all nervous	*somewhat nervous*	*moderately nervous*	*very nervous*

Since we last talked with you on (date of Follow-Up Interview 1)______ did you.

(1)speak with a family member to ask about your child s family history of diabetes? Y/N
(2)talk to someone who has a child with diabetes about your child' risk? Y/N

(3)talk to your pediatrician or other medical professional about your child's risk? Y/N
(4)call study personnel to discuss your child's risk? Y/N

(5)read a book or other literature on diabetes or diabetes risk? Y/N

(6)search the Internet for information on your child s risk? Y/N

Satisfaction with information received
Which of the following best describes the amount of information you received about the study
___You received too little information about the PANDA Part II study
___You received just the right amount of information about the PANDA Part II study

___You received too much information about the PANDA Part II study

At the present time, what are your plans with continuing with the PANDA study and having your child's blood tested again in the future?

___No, you are not considering continued participating at this time

___You're not sure right now if you are going to continue participating in the PANDA study

___Yes, you are definitely going to continue participating in PANDA Part II (make sure they have 800 number)

What are your thoughts on having had your child tested at birth for diabetes risk?

1 Glad I participated in the study
2 Not sure about our participation in the study
3 Wish I had not participated in the study

Thank you very much for your time tonight. If you have any questions about the study, please feel free to call Kerri North at 1-800-749-7424 ext. 27836, or locally at (352) 392-7836

APPENDIX H
PANDA GENERAL INFORMATION LETTER

About us ...What is the Panda Study? The Panda Study is a NIH and JDRF funded study, based at the University of Florida, which is focused on the genetic risks and development of Type I diabetes, formerly known as Juvenile Diabetes.

What is Type I Diabetes? Diabetes is a chronic metabolic disease. In the child with IDD (Insulin Dependent Diabetes) or Type I diabetes, the pancreas does not produce insulin, a hormone necessary to regulate blood sugar and to sustain life.

What causes Type I Diabetes? The disease occurs because the immune system attacks the cells in the pancreas that produce insulin. Without insulin the sugar in the blood cannot be used. It builds up in the bloodstream even while the body is starved for energy.

Is there a cure for Type I Diabetes? Not yet. There is no cure for Type I Diabetes. We are striving for a cure, but until a cure is found a person with Type I diabetes must take insulin injections daily to stay alive. Insulin, however, is not a cure. It is only a means of controlling the disease.

Our Research Plan The Panda Study involves two stages: initial screening for genetic risk and Antibody and immunological screening for those identified as being at genetic risk.

Panda I The genetic testing in newborns is performed in conjunction with the state mandated PKU/Metabolic screening test. The Panda screening will take an extra drop of blood and place it on filer paper before hospital discharge. For those who participate after leaving the hospital, a drop of blood is still required from a simple heel or finger stick.

Panda II The second part of the study is open to children who have participated in Panda I and are found to be at risk. At this point, about 2 to 3 teaspoons of blood is drawn from a vein to determine if the destruction of the insulin-producing cells in the pancreas has begun. We also offer the same screening as Panda I and II for the parents, and siblings of children in the Panda Study. For children identified as high risk, we hope to be able to enroll them in a prevention trial in the future.

Giving your consent for screening in the Panda Study Once you have decided to participate in our study you will be asked to sign an informed consent. Parents must give permission for children under 18 years of age. Along with this, an in depth questionnaire is filled out about your child's environment. We will also periodically contact you by phone for an update on your child's general health and well-being.

LIST OF REFERENCES

Ader, D.N., Sebring, A.R., Bhaskar, P., & Melamed, B. (1992). Information seeking and interactive videodisc preparation for third molar extraction. Journal of Oral and Maxillofacial Surgery, 50, 27-31

Abidin, R. (1986,1990). Parenting Stress Index. Charlottesville, VA: Pediatric Psychology Press.

Agre, P., Kurtz, R.C., & Krauss, B.J. (1994). A randomized trial using videotape to present consent information for colonoscopy. Gastrointestinal Endoscopy, 40(3), 271-276

Akerblom, H.K., Savilahti, E., Saukkonen, T.T., Paganus, A., Virtanen, S.M., Teramo, K., Knip, M., Ilonen, J., Reijonen, H., & Karjalainen, J. (1993). The case for elimination of cow's milk in early infancy in the prevention of Type I diabetes: the Finnish experience. Diabetes Metabolic Review, 9, 269-278

Alagna, S.W., Morokoff, P.J., Bevett, J.M., & Reddy, D.M. (1987). Performance of breast self-examination by women at high risk for breast cancer. Women & Health, 12(2), 29-46

Al-Jader, L.N., Goodchild, M.C., Ryley, H.C., & Harper, P.S. (1990). Attitudes of parents of cystic fibrosis children towards neonatal screening and antenatal diagnosis. Clinical Genetics, 38, 460-465

Alliance of Genetic Support Groups (1993). Available at www.geneticalliance.orgwww.geneticalliance.org. Accessed on 10/18/2003

American Academy of Pediatrics Committee on Bioethics. (2001). Ethical issues with genetic testing in pediatrics. Pediatrics, 107 (6), 1451-1455

American Psychological Association [APA] (2002). Ethical principles of psychologists and code of conduct. Available at: http://www.apa.org/ethics. Accessed on 11/02/2003

American Society of Human Genetics. (1996). ASHG Report: Statement on informed consent for genetic research. American Journal of Human Genetics, 59, 471-474

Axworthy, D., Brock, D.J.H., Bobrow, M., Marteau, T. (1996). Psychological impact of population-based carrier testing for cystic fibrosis: 3-year follow up. The Lancet, 347, 1443-1446

Baekkeskov, S , Landin O M Kristensen, J K., Srikanta, S Bruining, G J , & Mandrup-Poulsen, T et al , (1987) Antibodies to a Mr 64 000 human islet cell antigen precede the clinical onset of insulin dependent diabetes Journal of Clinical Investigation, 79, 926-934

Baroni, M.A., Anderson, Y E., & Mischler E.M (1997) Cystic fibrosis newborn screening Impact of early screening results on parenting stress Pediatric Nursing, 23(2), 143-145.

Baum, A., Friedman, A.L Zakowski, S G (1997) Stress and genetic testing for disease risk. Health Psychology, 16 (1) 8-19

Becker M H , & Maiman, L.A (1975) Sociobehavioral determinants of compliance with health and medical care recommendations Med Care, 13, 10-24

Bekker, H., Denniss, G., Modell, M , Bobrow, M., & Marteau, T (1994) The impact of population-based screening for carriers of cystic fibrosis. Journal of Medical Genetics, 31, 364-368

Bernhardt, B A., Geller G., Strauss, M Helzlsouer, K.J., Stefanek, M Wilcox, P M , Holtzmann, N.A (1997) Towards a model of informed consent process for BRCA1 testing A qualitative assessment of women's attitudes. Journal of Genetic Counseling, 6(2), 207-222

Biesecker, B.S , Boehnke M., Calzone, K. Markel, D , Garber J , Collings, F & Weber B (1992) Genetic counseling for families with inherited susceptibility to breast and ovarian cancer Journal of the American Medical Association, 269 1970-1974

Biesecker, B.B. Ishibe N, Hadley, D W Gumbarren, T.R., Kase, R., Lerman, C & Struewing, J.P (2000) Psychosocial factors predicting BRCA1/BRCA2 testing decisions in members of hereditary breast and ovarian cancer families. American Journal of Medical Genetics, 93, 257-263

Black, R.B (1982) Risk taking behavior: Decision making in the face of genetic uncertainty Social Work in Health Care, 7(1), 11-25

Bloch, M., Adams, S , Wiggins, S , Huggins, M & Hayden, M R. (1992) Predictive testing for Huntington's disease in Canada. The experience of those receiving an increased risk American Journal of Medical Genetics, 42, 499 507

Bodegard, G Fyro K & Larsson, A (1983) Psychological reactions in 102 families with a newborn who has a falsely positive screening test for congenital hypothyroidism. Acta Paediatr Scand Suppl , 304 1-21

Boland, C & Thompson N L. (1990) Effects of newborn screening of cystic fibrosis on reported maternal behavior Archives of Disease in Childhood, 65, 1240-1244

Bosompra, K., Flynn, B S., Ashikaga, T., Rairikar, C.J., Worden, J K., & Solomon, L.J (2000) Likelihood of undergoing genetic testing for cancer risk: a population based study Preventative Medicine, 30 155-166

Botkin, J R., Croyle, R.T , Smith, K.R., Baty, B.J , Lerman, C., Goldgar, D.E., Ward, J M Flick, B.J , & Nash, J E. A model protocol for evaluating the behavioral and psychosocial effects of BRCA1 testing. Journal of the National Cancer Institute (88), 13, 872-881

Bottorff, J L., Ratner, P.A., Johnson, J L. Lovato, C.Y., & Joab, S.A. (1998) Communication cancer risk information The challenges of uncertainty Patient Education and Counseling, 33, 67-81

Bowman, J E , (1993). Screening newborns for Duchene muscular dystrophy, British Medical Journal, 306 349

Bradley, D.M , Parsons, E.P , & Clarke, A.J (1993) Experience with screening newborns for Duschenne muscular dystrophy in Wales British Medical Journal, 306, 357-360

Brain, K. Gray, J., Norman, P , Parsons, E., Clarke, A., Rogers, C , Mansel, R., & Harper, P (2000) Why do women attend familial breast cancer clinics? Journal of Medical Genetics, 37, 197-202

Browner, C H Preloran, M & Press, N A (1996). The effects of ethnicity, education and an informational video on pregnant women's knowledge and decisions about a prenatal diagnostic screening test Patient Education and Counseling, 27, 135-146

Bruining G L , Molenaar J L , Grobee D , Hoffman, A., Scheffer, S L., Bruining, H.A deBruyn, A.M , Valkenburg, H A (1989) Ten year follow up study of islet cell antibodies and childhood diabetes mellitus. Lancet, i, 1100-1103

Bryant, G D., & Norman, G R. (1980) Expressions of probability words and numbers (letter) New England Journal of Medicine, 302, 411

Cameron, L.D , & Diefenbach, M. A. (2001) Responses to information about the psychosocial consequences of genetic testing for breast cancer susceptibility: Influences of cancer worry and risk perceptions Journal of Health Psychology, 6(1), 47-59

Carmichael, S L. K., Johnson, S B., Baughcum, Fuller, K. G , Taylor K. R., She, J X , & Schatz, D A. (2003) Newborn genetic screening for type 1 diabetes Maternal understanding of infant risk. Genetics in Medicine, 5(2), 77-83

Carmichael, S L K., Johnson, S B., & Tercyak, K (2000). Six-item short-form of the State-Trait Anxiety Inventory for use in a population undergoing genetic screening Unpublished manuscript, University of Florida.

Carver, C S , Pozo, C , Harris, S D , Noriega, V., Scheier, M.F., Robinson, D S , Ketcham, A.S , Moffat, F L , Jr & Clark, K C (1993) How coping mediates the effect of optimism on distress A study of women with early stage breast cancer Journal of Personality and Social Psychology, 65(2), 375 390

Carver, C S , Scheier, M F , & Weintraub, J K. (1989) Assessing coping strategies: a theoretically based approach. Journal of Personality and Social Psychology, 56, 267-283

Cassileth, B R , Zupkis, R.V , Sutton-Smith, K., and Marsh, V (1980) Informed consent why are its goals imperfectly realized? New England Journal of Medicine, 302, 896-900

Champion, V , Maraj, M , Hui, S , et al (2003) Comparison of tailored interventions to increase mammography screening in nonadherent older women. Preventive Medicine, 36 150-158

Chapman, E (2002) Ethical dilemmas in testing for late onset conditions Reactions to testing and perceived impact on other family members Journal of Genetic Counseling, 11 (5), 351-367

Chase, G.A, Faden, R.R. Holtzman, N.A., Chwalow, A.J., Leonard, C O., Lopes, C , and Quaid K (1983) Assessment of risk by pregnant women Implications for genetic counseling and education. Social Biology, 33(1-2), 57 64

Childs B , Gordis, L , Kaback, M.M , & Kazazian, H H (1976) Tay Sachs screening Social and psychological impact. American Journal of Human Genetics, 28, 550-558

Clark, S Bluman, L G Borstelmann, N., Regan, K.,Winer, E.P., Rimer, B K & Skinner, C S (2000) Patient motivation, satisfaction, and coping in genetic counseling and testing for BRCA1 and BRCA2 Journal of Genetic Counseling, 9(3), 219-235

Clarke, A. Parsons, E & Williams, A (1996) Outcomes and process in genetic counseling. Clinical Genetics, 50, 462-469

Clark, M A , Rakowski, W , Ehrich, B Rimer, B K , Velicer, W F Dube C.E., Pearlman, D N., Peterson, K K., Goldstein, M (2002) The effect of a stage-matched and tailored intervention on repeat mammography American Journal of Preventative Medicine, 22(1) 1-7

Clausen, H. Brandt, N.J Schwartz, M , & Skovby, F (1995) Psychosocial aspects in carrier screening for recessive disorders Nordisk Psykologi, 47(2), 81-103

Codori, A. & Brandt, J (1994). Psychological costs and benefits of predictive testing for Huntington's disease American Journal of Medical Genetics (Neuropsychiatric genetics, 54, 174-184

Codori, A M , Slavney, P.R., & Young C , et al (1997) Predictors of psychological adjustment to genetic testing for Huntington s Disease Health Psychology, 16, 36-50

Cohen, F Kearney K A., Zegans, L S., Kemeny M.E., Neuhaus, J.M., & Stites, D.P (1999). Differential immune system changes with acute and persistent stress for optimists vs. pessimists. Brain, Behavior, and Immunity, 13, 155-174

Collins F S , Green E D , Guttmacher A.E., & Guyer, M.S on behalf of the US National Human Genome Research Institute (2003) A vision for the future of genomics research Nature, 422(6934) 835-847

Croyle R.T & Lerman, C (1995) Psychosocial impact of genetic testing In. R.T Croyle, Ed. Psychosocial effects of screening for disease prevention and detection New York Oxford University Press.

Dahlquist, G , Blom, L Holmgren, G , Hogglof, B., Larsson, Y, Sterky, G (1985) The epidemiology of diabetes in Swedish children 0-14 years—A six years prospective study Diabetologia, 28, 802 808

Davis, J L Kawaguchi, Y Bennett, S T , Copeman, J B., Cordell, H.J., Pritchard, L.E , et al., (1994) A genome-wide search for human type 1 diabetes susceptibility genes. Nature, 37, 130-136.

Donovan K A , and Tucker D C (2000) Knowledge about genetic risk for breast cancer and perceptions of genetic testing in a sociodemographically diverse sample Journal of Behavioral Medicine, 23(1) 15-136

Duster T and Beeson, D (1997) Pathways and barriers to genetic testing and screening Molecular Genetics meets the "high risk family" Available from the Institute for the Study of Social Change at the University of California, Berkeley

Dutertre A E Barbier P Jonville, A.P , Pierre, F , & Berger C (1993) Parental opinions about biomedical research in children in Tours, France Dev Pharmacol Ther,20(1 2), 64-71

Edwards, A., and Elwyn, G (1999) How should effectiveness of risk communication to aid patients' decisions be judged? A review of the literature Medical Decision Making, 19, 428-434

Esplen, M J., Toner, B., Hunter, J , Glendon, G., Butler, K., & Field, B (1998) A group therapy approach to facilitate integration of risk information for women at risk for breast cancer Canadian Journal of Psychiatry, 43, 375-380

Edwards, S J., Lilford , R.J , Thornton, J , & Hewison, J (1998) Informed consent for clinical trials in search of the "best" method Social Science and Medicine, 47(11), 1825 1840

Evans, D G R., Blair, V , Greenhalgh, R., Hopwood, P, & Howell, A. (1994). The impact of genetic counseling on risk perception in women with a family history of breast cancer British Journal of Cancer, 70, 934 938

Evans, D G , Burnell, L D , Hopwood, P , & Howell, A (1993) Perception of risk in women with a family history of breast cancer British Journal of Cancer, 67, 612-634

Faden, R., & Beauchamp, T (1986) A history and theory of informed consent. New York Oxford University Press

Fanos, J H , & Johnson, J.P (1995) Barriers to carrier testing for adult cystic fibrosis sibs the importance of not knowing American Journal of Human Genetics, 59, 85-91

Fanos, J H , & Johnson, J P (1995) Perception of carrier status by cystic fibrosis siblings. American Journal of Human Genetics, 57, 431-438

Farmer, A.J., Levy, J C , & Turner, R.C (1999). Perceptions of risk of diabetes and anxiety following screening for Type II diabetes Diabetes, 48, Supplement 1

Farrell, P M , Kosorok, M.R , Loxeva, A , Shen, G , Kosick, R.E , Bruns, T , et al (1997) Nutritional benefits of neonatal screening for cystic fibrosis New England Journal of Medicine, 337, 936-939

Featherstone, K. & Donovan, J L (1998) Random allocation or allocation at random? Patients' perspectives of participation in a randomized controlled trial British Medical Journal, 17, 1177-1180

Fenton-May, J , Bradley, D M , Sibert, J R , Smith, R., Parsons, E P , Harper, P S., & Clarke A (1994) Screening for Duchenne muscular dystrophy Archives of Diseases in Childhood, 70(6), 551-2.

Flynn, J., Slovic, P , & Mertz, C.K. (1994). Gender, race and perception of environmental health risks. Risk Analysis, 14, 1101-1108

Freda, M.C., DeVore, N., Valentine-Adams, N., Bombard, A., & Merkatz, I.R. (1997) Informed consent for maternal serum alpha-fetoprotein screening in an inner city population How informed is it? Journal of Obstetrics, Gynecology and Neonatal Medicine, 27, 99-106

Frets, P.G., Duivenvoorden, H.J., Verhage, F., Niermeijer, M.F., Van de Berge, & S.M.M., Galjaard, H (1990) Factors influencing the reproductive decision after genetic counseling American Journal of Medical Genetics, 35, 492-502.

Friedman, L.C., Nelson, D.V., Baer, P.E., Lane, M., Smith, F.E., & Dworkin, R.J (1992) The relationship of dispositional optimism, daily life stress, and domestic environment to coping methods used by cancer patients. Journal of Behavioral Medicine, 15(2), 127-141

Fuller B.P., Kahn, M.J.E., Barr, P.A. Biesecker L., Crowley, E. Garber, J., Mansoura, M.K., Murphy P., Murray, J., Phillips, J. Rothenberg, K., Rothstein, M., Stopfer, J. Swergold, G., Weber, B., Collins, F.S., & Hudson, K.L (1999) Privacy in genetics research. Science, 285, 1359

Furr, L.A., & Seger R.E (1998). Psychological predictors of interest in prenatal genetic screening Psychological Reports, 82, 235-244

Garrud, P., Wood, M., & Stainsby L. (2001) Impact of risk information in a patient education leaflet. Patient Education and Counseling, 43, 301-304

Gillon, R. (2003). Ethics needs principles-four can encompass the rest-and respect for autonomy should be "first among equals" Journal of Medical Ethics, 29, 307-312

Gordon, C., Walpole, I., Zubrick, S.R., & Bower, C (2003) Population screening for cystic fibrosis Knowledge and emotional consequences 18 months later American Journal of Medical Genetics, 120A, 199-208

Green, J.M., Murton, F., & Statham, H. (1993) Psychosocial issues raised by a familial ovarian cancer register Journal of Medical Genetics, 30, 101-105

Gurm, H.S., & Litaker, D.G (2000) Framing procedural risks to patients: is 99% safe the same as a risk of 1 in 100? Academic Medicine, 75(8), 840-842

Hakel, M.D (1968) How often is often? American Psychologist, 23, 533-534

Hallowell, N. & Murton, F (1998). The value of written summaries of genetic consultations Patient Education and Counseling, 35, 27-24

Hallowell, N. Statham, H., Murton, F., Green, J., & Richards, M (1997) "Talking about chance" The presentation of risk information during genetic counseling for breast and ovarian cancer Journal of Genetic Counseling,35(6) 269-286

Hampton, M.L., Anderson, J., Lavizzo, B.S., & Bergman, A.B. (1974). Sickle cell "nondisease". A potentially serious public health problem. American Journal of Disease in Childhood, 128, 58-61

Harth, S.C., Johnstone, R.R., & Thong, Y.H. (1992). The psychological profile of parents who volunteer their children for clinical research: a controlled study. Journal of Medical Ethics, 18, 86-93

Harth, S.C., & Thong, Y.J. (1991). The social filter effect of informed consent in clinical research. Pediatrics, 87(4), 568-569

Howard Jones, N. (1982). Human experimentation in historical and ethical perspectives. Social Science and Medicine, 16, 1429-1448

Hughes, C., Gomez-Caminero, A., Benkendorf, J., Kerner, J., Issacs, C., Barter, J., & Lerman, C. (1997). Ethnic differences in knowledge and attitudes about BRCA1 testing in women at increased risk. Patient Education and Counseling, 32, 51-62

Institute of Medicine (IOM). Committee on Assessing Genetic Risks. (1994). Assessing genetic risks: Implications for health and social policy. L.B. Andrews, J.E. Fullarton, N.A. Holtzman, and A.G. Motulsky (Eds.) Washington, D.C.: National Academy Press

Janis, I. L., & Feshback, S. (1953). Effects of fear-arousing communications. Journal of Abnormal and Social Psychology, 48, 78-92

Jimison, H.B., Sher, P.P., Appleyard, R., and LeVernois, Y. (1998). The use of multimedia in the informed consent process. Journal of the American Medical Information Association, 5(3), 245-256

Johnson, S.B. (2001). Screening programs to identify children at risk for diabetes mellitus: Psychological impact on children and parents. Journal of Pediatric Endocrinology & Metabolism, 14, 653-659

Johnson, S.B., Baughcum, A.E., Carmichael, S.L.K., She, J.X., & Schatz, D. (in press). Diabetes Care.

Johnson, S.B., & Carmichael, S.L.K. (2000). At-risk for diabetes: Coping with the News. Journal of Clinical Psychology in Medical Settings, 7(1), 69-78

Johnson, S.B., Riley, W.J., Hansen, C.A., & Nurick, M.A. (1990). Psychological impact of islet cell antibody screening. Diabetes Care, 13, 93-97

Johnson, S B & Tercyak, K. (1995) Psychological impact of islet cell antibody screening for IDDM on children, adults, and their family members Diabetes Care, 18(10), 1370-1372

Jones, J.H. (1981) Bad blood, New York Free Press.

Kash, K.M , Holland, J C , Halper, M S , & Miller, D G (1992) Psychological distress and surveillance behaviors of women with a family history of breast cancer Journal of the National Cancer Institute, 84 24-30

Kash, K.M., Holland, J C., Osborne, M.P & Miller, D.G (1995) Psychological counseling strategies for women at risk of breast cancer Journal of the National Cancer Institute, 17, 73-79

Kash, K.M., Ortega-Verdejo, K., Dabney, M K Hollands, J C , Miller, D G & Osbourne, M P (2000) Psychosocial aspects of cancer genetics women at high risk for breast and ovarian cancers Seminars in Surgical Oncology, 18, 333-338

Kenen, R., Smith, A.C.M., Watkins C , & Zuber-Pittore, C (2000) To use of not to use the prenatal genetic technology/worry conundrum Journal of Genetic Counseling, 9(3), 203-217

Kenney, R.M (1981) Between never and always The New England Journal of Medicine, 305, 1097-1098

Kerem, B S., Rommens, J M Buchanan, J A , Markiewicz, D Cox T K., Chakravarto, A., Buchwald, M., & Tsui, L. (1989) Identification of the cystic fibrosis gene Genetic analysis. Science, 245, 1116-1120

Kessler, S (1988) Invited essay on the psychological aspect of genetic counseling V Preselection A family coping strategy in Huntington s disease American Journal of Medical Genetics, 31, 617-621

Kessler, S (1989) Psychological aspects of genetic counseling VI A Critical Review of the literature dealing with education and reproduction. American Journal of Medical Genetics, 34, 340-353

Knip, M.(1998) Prediction and prevention of type 1 diabetes. Pediatric Supplement, 425, 54-62

Kong, A Barnett, G O , Mosteller, F , & Youtz, C (1986). How medical professional evaluate expressions of probability The New England Journal of Medicine, 315,740-744

Kwon, C , & Farrell, P M (2000) The magnitude and challenge of false-positive newborn screening test results Archives of Pediatric and Adolescent Medicine, 154 (7), 714-718

Kyvik, K. O , et al. (1995) Concordance rates of insulin dependent diabetes mellitus: a population based study of young Danish twins [see comments] British Medical Journal, 311, 913-917

LaPorte, R., M , et al , (1999) Prevalence and incidence of insulin-dependent diabetes. In (Ed. 2nd), Diabetes in America (pp 37-14) Bethesda, MD National Diabetes Data Group, NIH

Landin-Olsson, M , Christie, M R., Standl, E., Ziegler, A., Blom, L , Sundvist, G., Nystrom, L. et al (1992). Predictive value of islet cell and insulin autoantibodies for Type I (insulin-dependent) diabetes mellitus in a population-based study of newly-diagnosed diabetic and matched control children. Diabetologia, 35, 1068-1073

Lawson, S L & Adamson, H M (1995) Informed consent readability Subject understanding of 15 common consent form phrases IRB 16-19

Lazarus, R S , & Folkman, S (1984) Stress, appraisal, and coping. New York Springer

Lerman, C (1997) Translational behavioral research in cancer genetics Preventative Medicine, 26 S65-S69

Lerman, C , Biesecker, B Benkendorf J L , Kerner, J , Gomez-Caminero, A., Hughes, C , & Reed, M M (1997) Controlled trial of pre test education approaches to enhance informed decision-making for BRACA1 gene testing. Journal of the National Cancer Institute, 89(2), 148-157

Lerman, C , Daly M Sands, C Balshem, A. Lustbader, E , Heggan, T., Goldstein, L James, J & Engstrom P (1993) Mammography adherence and psychological distress among women at risk for breast cancer Journal of the National Cancer Institute, 85(13) 1974-1080

Lerman, C Kash, K. & Stefanek, M (1994) Younger women at increase risk for breast cancer: Perceived risk, psychological well-being, and surveillance behavior Monographs: National Cancer Institute, 15, 171-176

Lerman, C , Lustbader, E , Rimer, B Daly M Miller, S , Sands, C & Balshem, A. (1995) Effects of individualized breast cancer risk counseling A randomized trial Journal of the National Cancer Institute, 87 286-292

Lerman, C , Miller, S M., Scarborough R , Hanjani, R., Nolte, S & Smith, D (1991) Adverse psychological consequences of positive cytologic cervical screening. American Journal of Obstetrics and Gynecology, 165, 658-652

Lerman, C., Narod, S., Schulman, K., Hughes, C., et al. (1996). BRCA1 testing in families with hereditary breast-ovarian cancer. A prospective study of patient decision making and outcomes. Journal of the American Medical Association, 275, 1885-1892

Lerman, C., Rimer, B., Blumberg, B., Cristinzio, S., Engstrom, P.F., MacElwee, N., O'Connor, K., & Seay, J. (1990). Effects of coping style and relaxation on cancer chemotherapy side effects and emotional responses. Cancer Nursing, 13(5), 308-315.

Lerman, C., Schwartz, M.D., Miller, S.M., Daly, M., Sands, C., & Rimer, B.K. (1996). A randomized trial of breast cancer risk counseling: Interacting effects of counseling, educational level, and coping style. Health Psychology, 15(2), 75-83

Lerman, C., Seay, J., Balshem, A., & Audrain, J. (1995). Interest in genetic testing among first-degree relatives of breast cancer patients. American Journal of Medical Genetics, 57, 385-392

Leventhal, H., Nerenz, D.R., & Steele, D.J. (1984). Illness representations and coping with health threats. In A. Baum, S.E. Taylor, J.E. Singer et al. (Eds.), Handbook of psychology and health, Vol. 4. pp. 219-252. New York: Springer Verlang

Lipkus, I.M., Rimer, B.K., Halabi, S., & Strigo, T.S. (2000). Can tailored interventions increase mammography use among HMO women? American Journal of Preventive Medicine, 18(1), 1-10

Lippman-Hand, A., and Fraser, F.C. (1979a). Genetic counseling: parents' responses to uncertainty. Birth Defects Original Article Series, 15(5c), 325-339

Lippman-Hand, A., and Fraser, F.C. (1979b). Genetic counseling: provision and reception of information. American Journal of Medical Genetics, 3, 113-127

Litt, M.D., Tennen, H., Affleck, G., & Klock, S. (1992). Coping and cognitive factors in adaptation to in vitro fertilization failure. Journal of Behavioral Medicine, 15(2), 171-187

Llewellyn-Thomas, H.A. (1995). Patients' health-care decision making: a framework for descriptive and experimental investigations. Medical Decision Making, 15(2), 101-106.

Lobel, M., Yali, A.M., & Zhu, W. (1998). Emotional reactions to the stress of high-risk pregnancy: The role of optimism and coping. Annals of Behavioral Medicine, 20, 29

Lucidarme, N., Domenges-Muriel, W., Castro, D., Czernichow, P., & Levy Marchal, C. (1998). Appraisal and implications of predictive testing for insulin-dependent diabetes mellitus. Diabetes and Metabolism, 24, 550-553

Ludwick-Rosenthal R., & Neufeld R.W. (1993). Preparation for undergoing an invasive medical procedure: interacting effects of information and coping style. Journal of Consulting and Clinical Psychology, 61(1), 156-164.

Mapes, R.E.A. (1979). Verbal and numerical estimates of probability in therapeutic contexts. Social Science and Medicine, 13A, 277-282.

Markel, H. (1992). The stigma of disease: Implications of genetic screening. The American Journal of Medicine, 93, 209-215.

Markel, D.S., Young, A.B., & Penney, J.B. (1987). At risk persons' attitude toward presymptomatic and prenatal testing of Huntington disease in Michigan. American Journal of Medical Genetics, 26, 295-305.

Marteau, T.M., & Bekker, H. (1992). The development of a six-item short-form of the state scale of the Spielberger State-Trait Anxiety Inventory (STAI). British Journal of Clinical Psychology, 31(3), 301-306.

Marteau, T.M., Kidd, J., Cook, R., Michie, S., Johnston, M., Slack, J., & Shaw, R.W. (1991). Perceived risk not actual risk predicts uptake of amniocentesis. British Journal of Obstetrics and Gynaecology, 98, 282-286.

Marteau, T.M., & Michie, S. (1995). Letters: A positive screening test for an untreatable condition provides psychological relief from uncertainty. British Journal of Medicine, 311, 58-59.

McNeil, T.F., Sveger, T., & Thelin, T. (1988). Psychosocial effects of screening for somatic risk. The Swedish alpha-1-antitrypsin experience. Thorax, 43, 505-507.

Meissen, G.J., Myers, R.H., Mastromauro, C.A., Koroshetz, W. (1988). Predictive testing for Huntington's disease with use of a linked DNA marker. New England Journal of Medicine, 318(9), 535-542.

Meissen, G.J. & Berchek, R.L. (1987). Intended use of predictive testing by those at risk for Huntington disease. American Journal of Medical Genetics, 26, 283-293.

Melville, N.A. (2001). Public struggles with genetics 101. Health Scout News, May 27, 2001. Available at http://dailynews.yahoo.com/h/hsn/20010528/hl/public_struggles_with_genetics_101_1.html. Accessed July 1, 2001.

Merz, J.F., Druzdzel, M.J., & Mazur, D.J. (1991). Verbal expressions of probability in informed consent litigation. Medical Decision Making, 11, 273-281.

Michie, S Bobrow, M , & Marteau, T M (2001) Predictive genetic testing in children and adults A study of emotional impact Journal of Medical Genetics, 38 519-526

Michie, S., French, D , Allanson, A , Bobrow, B, & Marteau, T (1997) Information recall in genetic counselling: a pilot study of its assessment. Patient Education and Counseling, 32, 93-100

Miller, R. & Willner, H S (1974) Sounding Board. The Two-Part Consent Form The New England Journal of Medicine, 964-966

Miller, S M (1980) When is a little information a dangerous thing? Coping with stressful events by monitoring versus blunting. In S Levine & H Ursin (Eds.), Coping and Health (pp 45-69) New York Plenum.

Miller, S M (1983) Monitoring and blunting Validation of a questionnaire to assess style of information seeking under threat. Journal of Personality and Social Psychology, 52, 345-353

Miller S M., Combs, C., & Stoddard, E. (1989) Information, coping and control in patients undergoing surgery and stressful medical procedures Steptoe, A and Appels, A (Eds) Stress, personal control and health (pp 107-130) Oxford, England John Wiley & Sons

Miller, S M & Mangan, C (1983) Interacting effect of information and coping style in adapting to gynecologic stress should the doctor tell all? Journal of Personality and Social Psychology, 45 223-236

Modell, B (1992). Ethical aspects of genetic screening Annals of Medicine, 24, 549-555

Moseley R., Allen, B., Van McCrary S. Dewar M., Crandall, L A & Ostrer H (1995) The ethical, legal, and social implications of predictive genetic testing for health, life and disability insurance policy analysis and recommendations A report from the Human Genome Insurance Project, Medical Humanities Program, University of Florida College of Medicine

Nako, M.A., & Axelrod, S (1983) Numbers are better than words Verbal specifications of frequency have no place in medicine The American Journal of Medicine, 74, 1061-1065

National Center for Education Statistics (2001) Accessible at http://nces.ed.gov/ Accessed November 15 2003

National Human Genome Research Institute Ethical, Legal and Social Implications (ELSI) Research Program (2003) Available at http://www.nhgri.nih.gov/10001618 Accessed November 15, 2003

Nisbett, R., & Ross L (1980) Human inference. Strategies and shortcomings of social judgment. Englewood Cliffs, JN Prentice-Hall

O'Connor B J V (1996) Predictive genetic testing for cystic fibrosis carrier status the role of health beliefs and coping style Masters Abstracts International, 36(2), 0630

Oliner, S P & Oliner P M (1988) The altruistic personality New York. Free Press

Ormond, K.E , Pergament, E , and Fine, B A (1996) Pre-screening education in multiple marker screening programs the effect on patient anxiety and knowledge. Journal of Genetic Counseling, 5(2), 69 80

Pepper S & Prytulak, L S (1974) Sometimes frequently means seldom context effects in the interpretation of quantitative expressions Journal Res Personality, 8, 95-101

Peters, T (1993) Genomore project forces new look at ethics, law Forum for Applied Research and Public Policy, Fall, 5-13

Project Policy Report (2003) Executive Summary Genome technology and reproduction. Values and public policy Available at http://www.sph.umich.edu/genpolicy/initialprojectsummary/policeyreport.pdf Accessed on 11/02/2003

Quaid, K Brandt, J Faden, R. & Folstein, S (1989) Knowledge, attitude, and the decision to be tested for Huntington's Disease Clinical Genetics, 36, 431-438

Rewers, M , Bugawan, T L , Norris, J M Blair, A , Beaty, B Hoffman, M McDuffie, R S Jr Hamman, R.F Klingensmith, G , Eisenbarth, G.S , & Erlich. (1996) Newborn screening for HLA markers associated with IDDM Diabetes Autoimmunity Study in the Young (DAISY) Diabetologia, 39, 807 812

Richards, M (1996) Families, kinship, and genetics. In T Marteau & M Richards (Eds), The troubled helix Social and psychological implications of the new human genetics. Great Britain Cambridge University Press, pp 249-273

Riley, W J., McClaren, N K., Krischer, J Spillar, R P Silverstein, J H., Schatz, D.A , Schwartz, S et al (1990) A prospective study of the development of diabetes in relatives of patients with insulin dependent diabetes New England Journal of Medicine, 323, 1167 1172

Rimer, B.K., & Glassman, B. (1999). Is there a use for tailored print communications in cancer risk communications? Journal of the National Cancer Institute Monographs, 25, 140-148

Rimer, B.K., Halabi, S., Skinner, C.S., Kaplan, E.B., Crawford, Y., Samsa, G.P., Strigo, T.S., & Lipkus, I.M. (2001). The short-term impact of tailored mammography decision-making interventions. Patient Education and Counseling, 43, 269-285

Rimer, B.K., Halabi, S., Skinner, C.S., et al. (2002). Effects of mammography decision making intervention at 12 and 24 months. American Journal of Preventive Medicine, 22, 247-257

Roll, U., Christie, M.R., Standl, E., & Ziegler, A.G. (1994). Associations of anti-GAD antibodies with islet cell antibodies and insulin autoantibodies in first-degree relatives of Type I diabetic patients. Diabetes, 43, 154-160

Rosenstock, I.M. (1974). The health belief model and preventative health behaviors. Health Education Monographs, 2, 254-386

Rothenberg, M., & Sills, E. (1968). The PKU anxiety syndrome. Journal of the American Academy of Child Psychiatry, 7, 689-692

Rothert, M.L., Holmes-Rovner, M., Rovner, D., Kroll, J., Breer, L., Talarczyk, G., Schmitt, N., Padonu, G., & Wills, C. (1997). An educational intervention as decision support for menopausal women. Research in Nursing & Health, 20, 377-387

Rowley, P.T., Loader, S.L., Sutera, C.J., Walden, M., & Kozyra, A. (1991). Prenatal screening of hemoglobinopathies. III. Applicability of the health belief model. American Journal of Human Genetics, 48, 452-459

Rushton, J.P., Chrisjohn, R.D., & Fekken, G.C. (1981). The altruistic personality and the self-report altruism scale. Personality and Individual Differences, 2, 293-302.

Sagi, M., Shiloh, S., & Cohen, T. (1992). Application of the health belief model in a study on parent' intentions to utilize prenatal diagnosis of cleft lip and/or palate. American Journal of Medical Genetics, 44, 326-333

Schatz, D., Krischer, J., Horne, G., Riley, W., Spillar, R., Silverstein, J., Winter, W., Muir, A., Derovanessian, D., Shah, S., Malone, J., & Maclaren, N. (1994). Islet cell antibodies predict insulin-dependent diabetes in United States school age children as powerfully as in unaffected relatives. Journal of Clinical Investigation, 93, 2403-2407

Schatz, D., Muir, A., Fuller, K., Atkinson, M., Crockett, S., Hsiang, H., Winter, B., Ellis, T., Taylor, K., Sattes, C., Dukes, M., Fang, Q., Clare-Salzer, M., & She, J. (2000). Prospective assessment in newborns for diabetes autoimmunity (PANDA): A newborn screening program in the general population of Florida. Abstracts from the 60th scientific sessions of the American Diabetes Association (ADA), A67

Scheier, M.F., & Carver, C.S. (1985). Optimism, coping, and health: Assessment and implications of generalized outcome expectancies. Health Psychology, 4(3), 219-247

Scheier, M.F., Matthews, K.A., Owens, J.F., Magovern, G.J., Lefebvre, R.C., Abbott, R.A., & Carver, C.S. (1989). Dispositional optimism and recovery from coronary artery bypass surgery: the beneficial effects on physical and psychological well-being. Journal of Personality and Social Psychology, 57(6), 1024-1040

Scheier, M.F., Weintraub, J.K., & Carver, C.S. (1986). Coping with stress, divergent strategies of optimists and pessimists. Journal of Personality and Social Psychology, 51(6), 1257-1264

Schwartz, M.D., Lerman, C., Miller, S.M., Daly, M., & Masny, A. (1995). Coping disposition, perceived risk, and psychological distress among women at increased risk for ovarian cancer. Health Psychology, 14(3), 232-235

Segerstrom, S.C., Taylor, S.E., Kemeny, M.E., & Fahey, J.L. (1998). Optimism is associated with mood, coping, and immune changes in response to stress. Journal of Personality and Social Psychology, 74, 1646-1655

Shaw, N.J., & Dear, P.R.F. (1990). How do parents of babies interpret qualitative expressions of probability? Archives of Disease in Childhood, 65, 520-523

Shiloh, S., & Sagi, M. (1989). Effect of framing on the perception of genetic recurrence risks. American Journal of Medical Genetics, 33, 130-135

Shiloh, S., & Saxe, L. (1989). Perception of risk in genetic counseling. Psychological Health, 3, 45-61

Shoda, Y., Mischel, W., Miller, S.M., Diefenbach, M., Daly, M.B., & Engstrom (1998). Psychological interventions and genetic testing: Facilitating informed decisions about BRCA1/2 cancer susceptibility. Journal of Clinical Psychology in Medical Settings, 5(1), 3-17

Silva, M.C., & Sorrell, J.M. (1988). Enhancing comprehension of information for informed consent: a review of empirical research. IRB, 10(1), 1-5

Silverman, W.A. (1989) The myth of informed consent in daily practice and in clinical trials Journal of Medical Ethics, 15, 6-11

Slovic, P., Fischhoff, B., and Lichtenstein, S (1982) Response modes, framing, and information—processing effects in risk assessment In R Hogarth (Ed), New directions for methodology of social and behavioral science, Question framing and response consistency. San Francisco Jossey Bass p 21-36

Sobel, S K., & Cowan, D B (2000) Impact of genetic testing for Huntington Disease on the family system. American Journal of Medical Genetics, 90, 49-59

Sorenson, J R., Scotch, N.A., Swazey, J P., Wertz, D C., & Heeren, T C (1987) Reproductive plans of genetic counseling clients not eligible for prenatal diagnosis American Journal of Medical Genetics, 28, 345 352.

Speilberger CD, Gorsuch RL, & Lushene R (1980) Test manual for the State-Trait Anxiety Inventory Palo Alto, CA, Consulting Psychologists Press.

Speilberger C D (1973) Test manual for the State-Trait Anxiety Inventory for Children Palo Alto, CA, Consulting Psychologists Press, 1973

Sweet, K.M. Willis, S K., Ashida, S., & Westman, J.A (2003) Use of fear-appeal techniques in the design of tailored cancer risk communication messages Implications for healthcare providers. Journal of Clinical Oncology, 21(17), 3375-3376

Szilagyi, P G Bordley, C., Vann, J C , Chelminski, A., Kraus, R.M , Margolis P A , & Rodewald, L E. (2000) Effect of patient reminder/recall interventions on immunization rates. A review Journal of the American Medical Association, 284, (14), 1820-1827

Taub H A., Baker M T & Sturr, J.F (1986) Informed consent for research effects of readability, patient age, and education. Journal of the American Geriatrics Society, 34, 601-606

Taylor, S E., Kemeny M.E., Aspinwall, L.G , Schneider, S G., Rodriquez, R. & Herbert, M (1992) Optimism, coping, psychological distress, and high-risk sexual behavior among men at risk for acquired immunodeficiency syndrome (AIDS) Journal of Personality and Social Psychology, 63(3), 460-473

Tercyak, K.P , Johnson, S B , Roberts S. & Cruz, A (2001).Psychological response to prenatal genetic counseling and amniocentesis Patient Education and Counseling, 43(1), 73-84

Tercyak, K.P., Johnson, S.B., & Schatz, D.A. (1998). Patient and family perceptions of the use of subcutaneous insulin to prevent IDDM: Implications for current and future prevention trials. Journal of Diabetes and its Complications, 12, 279-285.

Tercyak, K.P., Streisand, R., Peshkin, B., & Lerman, C. (2000). Psychosocial impact of predictive testing for illness on children and families: Challenges for a new millennium. Journal of Clinical Psychology in Medical Settings, 7, 55-68.

Tibben, A., Duivenvoorden, H.J., Vegter-van der Vlis, M., Niermeijer, M.F., Frets, P.G., van der Kamp, J.J.P., et al. (1993a). Presymptomatic DNA Testing for Huntington's Disease: Identifying the need for psychosocial intervention. American Journal of Medical Genetics, 48, 137-144.

Tibben, A., Frets, P.G., van der Kamp, J.J.P., Niermeijer, M.F., Vegter-van der Vlis, M., Skraastad, M.I., Roos, R.A.C., Rooymans, H.J.M., van Ommen, G.J.B., & Verhage, F. (1993b). On attitudes and appreciation 6 months after predictive DNA testing for Huntington Disease in the Dutch Program. American Journal of Medical Genetics, 48, 103-111.

Tibben, A., Vegter-van der Vlis, M., Skraastad, M.I., Frets, P.G., van der Kamp, J.J.P., Niermeijer, M.F., van Ommen, G.B., Roos, R.A.C., Rooijmans, H.G.M., Stronks, D., & Verhage, F. (1992). DNA-Testing for Huntington's Disease in the Netherlands: A retrospective study on psychosocial effects. American Journal of Medical Genetics, 44, 94-99.

Tluczek, A., Mischler, E.H., Bowers, B., Peterson, N., Morris, M.E., Farrell, P.M., Bruns, W.T., Colby, H., McCarthy, C., Fost, N., & Carey, P. (1991). Psychological impact of false-positive results when screening for cystic fibrosis. Pediatric Pulmonology Supplement, 7, 29-37.

Tucker, C., & Pedersen, T. (personal communication, 2000). Patients want more culturally sensitive health care. Manuscript in preparation. University of Florida Department of Psychology.

Tversky, A., & Kahnemann, D. (1981). The framing of decisions and the psychology of choice. Science, 211, 453-458.

Tymchuk, A.J. (1998). Informing for consent: concepts and methods. Canadian Psychology, 38(2), 55-75.

Valanis, B., Whitlock, E.E., Mullooly, J., Vogt, T., Smith, S., Chen, C., & Glasgow, R.E. (2003). Screening rarely screened women: time-to-service and 24-month outcomes of tailored interventions. Preventive Medicine, 37, 442-450.

VanMcCrary, S., Allen, B. Moseley, R., Crandall, L.A. Ostrer, H., Curry, R.W., Dewar, M.A., & Nye, D. (1993). Ethical and practical implications of the human genome initiative for family medicine. Archives of Family Medicine, 2, 1158-1163

Van Stuijvenberg, Suur, M.H., de Vos, S., Tjiang, C.H., Steyerberg, E.W., Derksen-Lubsen, G., & Moll, H.A. (1998). Informed consent, parental awareness, and reasons for participating in a randomised controlled study. Archives of Diseases of Childhood, 79, 120-125

Van 't Spijker, A., & ten Kroode, H.F.J. (1997). Psychological aspects of genetic counselling. A review of the experience with Huntington's disease. Patient Education and Counseling, 32, 33-40

Vernon, S.W., Vogel, V.G., Halabi, S., & Bondy, M.L. (1993). Factors associated with perceived risk of breast cancer among women attending a screening program. Breast Cancer Research and Treatment, 28, 137-144

Verheggen, F., Jonkers, R., & Kok, G. (1996). Patient's perceptions of informed consent and the quality of information disclosure in clinical trials. Patient Education and Counseling, 29, 147-153

Waggoner, W.C., & Maye, D.M. (1995). Who understands? A survey of 25 words or phrases commonly used in proposed clinical research consent forms. IRB, 17(1), 6-9

Wagner, A., Tibben, A. Bruining, G.J., Aanstoot, H.J., Tiems, I., Blondeau, M.J., & Niermeijer, M.F. (1995). Preliminary experience with predictive testing for insulin-dependent diabetes mellitus. Lancet, 246, 380-381

Wald, N.J., & Morris, J.K. (1998). Neonatal screening for cystic fibrosis. British Journal of Medicine, 316, 404-405

Walters, D., Wilcken, B., Irwig, L., Simpson, J., Van Asperen, P., & Gaskin, K. (1999). Clinical outcomes from newborn screening for cystic fibrosis in New South Wales. Arch Dis Child Fetal Neonatal, 80, 1-7

Wardle, J., Collins, W., Pernet, A., Whitehead, M., Bourne, H., & Campbell, S. (1995). Psychological impact of screening for familiar ovarian cancer. Journal of the National Cancer Institute, 86, 653-657

Watson, E., Mayall, E., Lamb, J., Chapple, J., & Williamson, B. (1992). Psychological and social consequences of community carrier screening programmes for cystic fibrosis. Lancet, 340, 217-220

Weinstein, N.D. (1984). Why it won't happen to me: perceptions of risk factors and susceptibility. Health Psychology, 3(6), 431-457

Weinstein, N.D (1988) The precaution adoption process Health Psychology, 7 355-386

Wells, S.A. Jr., & Skinner, M.A. (1998) Prophylactic thyroidectomy, based on direct genetic testing, in patients at risk for the multiple endocrine neoplasia type 2 syndromes. Experiences in Clinical Endocrinology & Diabetes, 1061 29-24

Wertz, D C., Sorenson, J.R. & Heeren, T C (1986) Clients' interpretation of risks provided in genetic counseling. American Journal of Human Genetics, 39, 253-264.

Wertz, D C Fanos, J.H. & Reilly P R. (1994) Genetic testing for children and adolescents Who decides? Journal of the American Medical Association, 272(11), 875-881

Wexler, N S (1984) Huntington's disease and other late onset genetic disorders In A E.H Emer & L M Pullen (Eds.), Psychological aspects of genetic counseling London Academic Press.

Weston, J , Hannah, M., and Downes, J (1997) Evaluating the benefits of a patient information video during the informed consent process Patient Education and Counseling, 30, 239-245

Wilken, B & Travert, G (1999) Neonatal screening for cystic fibrosis: present and future Acta Paediatrica Supplement, 432, 33 35

Wilken, B , Towns, S.J., & Mellis, C M. (1983) Diagnostic delay in cystic fibrosis. Lessons from newborn screening Archives of Diseases in Childhood, 58, 863-866

Wilkenson, C., Jones, J M., & McBride, J (1990) Anxiety caused by abnormal result of cervical smear test a controlled trial British Journal of Medicine, 300, 440

Yu, M., Norris, J., Mitchell C Butler-Simon, N., Groshek, M., Follansbee D , Erlich, H., Rewers, M., & Klingensmith, G (1999) Impact on maternal parenting stress of receipt of genetic information regarding risk of diabetes in newborn infants American Journal of Medical Genetics, 86, 219-226

Zeesman, S., Clow, C L., Carter L., & Scriver, C.R. (1984) A private view of heterozygosity Eight year follow up study on carriers of the Tay-Sachs detected by high school screening in Montreal American Journal of Medical Genetics, 18, 769-778

Ziegler A G Hummel M Schenker, M , & Bonifacio E (1999) Antibody appearance and risk for development of childhood diabetes in offspring of parents with Type I diabetes The 2-year analysis of the German BABYDIAB study. Diabetes, 48, 460-468

BIOGRAPHICAL SKETCH

Stacy Koser Carmichael obtained a Bachelor of Science degree in biology with a double major in psychology from Buena Vista University in Storm Lake, Iowa, in January of 1996. Stacy enrolled at the University of Florida in September of 1996 for doctoral work in clinical and health psychology, concentrating on child and pediatric psychology. Stacy completed her pre-doctoral internship at the A.I. duPont Hospital for Children in Wilmington, Delaware, in the 2002-2003 academic year. She is currently completing a two-year psychology fellowship in the Department of Medical Humanities and Social Sciences at the Florida State University College of Medicine. Stacy lives with her son Jaden in Tallahassee, Florida.

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy

Suzanne Bennett Johnson

Suzanne Bennett Johnson, Chair
Professor of Clinical and Health Psychology

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy

Desmond Schatz
Professor of Pediatrics

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy

Steph Boggs

Stephen Boggs
Associate Professor of Clinical and Health Psychology

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy

Gary Geffken
Associate Professor of Clinical and Health Psychology

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy

Fonda Eyler
Professor of Psychology

This dissertation was submitted to the Graduate Faculty of the College of Health Professions and to the Graduate School and was accepted as partial fulfillment of the requirements for the degree of Doctor of Philosophy

December 2003

Robert G Frank

Dean, College of Health Professions

Dean, Graduate School

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy

Suzanne Bennett Johnson

Suzanne Bennett Johnson, Chair
Professor of Clinical and Health Psychology

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy

Desmond Schatz
Professor of Pediatrics

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy

Steph Boggs

Stephen Boggs
Associate Professor of Clinical and Health Psychology

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy

Gary Geffken
Associate Professor of Clinical and Health Psychology

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy

Fonda Eyler
Professor of Psychology

This dissertation was submitted to the Graduate Faculty of the College of Health Professions and to the Graduate School and was accepted as partial fulfillment of the requirements for the degree of Doctor of Philosophy

December 2003

Robert G Frank

Dean, College of Health Professions

Dean, Graduate School